



DOCTOR OF CLINICAL PSYCHOLOGY (DCLINPSY)

Doctorate in Clinical Psychology: Main Research Portfolio.

1) Self-Report Cognitive Factors Maintaining A Failure To Speak In Selective Mutism; 2) Sex and Relationships Education for Individuals with Cystic Fibrosis: A Service-Directed Approach; 3) The Relationship between Challenging Behaviour and Social Story Interventions: A Pilot Study in a Naturalistic Setting.

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Research Portfolio Submitted in Part Fulfilment of the requirements for the Degree of Doctorate in Clinical Psychology

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Doctorate in Clinical Psychology

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[May 2017]

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Table of Contents

LIST OF APPENDICES.....	8
LIST OF TABLES.....	10
LIST OF FIGURES.....	12
ABSTRACTS.....	13
CRITICAL REVIEW OF THE LITERATURE: Self-Report Cognitive Factors Maintaining A Failure To Speak In Selective Mutism.....	16
INTRODUCTION.....	17
Description and History of Selective Mutism.....	17
Theoretical Understanding of Selective Mutism.....	18
Intervention for Selective Mutism.....	19
Rationale	20
METHOD.....	21
Review Design.....	21
Inclusion Criteria.....	23
Participants.....	23
Intervention and outcome.....	23
Study design.....	23
Exclusion Criteria.....	23
Participants.....	24
Intervention and outcome.....	24
Study design.....	24
Information Sources.....	24
Data Collection and Analysis.....	26
Study Selection.....	26
Data Extraction Process.....	27
RESULTS.....	37
Quality of Studies.....	37
Demographic and Methodological Issues.....	37
Characteristics of Sample.....	37

Characteristics of Participants.....	38
Content Analysis.....	38
Categories for content analysis.....	39
<i>Failure to speak due to anxiety</i>	39
<i>Beliefs about control over speaking</i>	39
<i>Beliefs about mutism as protective</i>	40
<i>Negative self-perception</i>	41
<i>Expectations of others</i>	41
<i>Perception of separate identity as mute</i>	41
<i>Beliefs around the reason for mutism being unknown</i>	42
DISCUSSION.....	43
Agreements and Disagreements with Other Papers.....	44
Limitations and Biases.....	45
CONCLUSIONS AND CLINICAL IMPLICATIONS.....	46
REFERENCES.....	48
SERVICE IMPROVEMENT PAPER: Sex and Relationships Education for Individuals with Cystic Fibrosis: A Service-Directed Approach.....	53
INTRODUCTION.....	54
BACKGROUND LITERATURE.....	54
Key Sexual and Reproductive Health (SRH) Issues in CF.....	54
SRH Provision.....	55
Context of Local Service Provision.....	56
Aims.....	57
Research Questions.....	57
Design.....	58
METHOD.....	58
Procedure.....	58
Phase 1: Service consultation and scoping.....	58
Phase 2: Questionnaire design.....	58
Phase 3: Survey.....	59

<i>Recruitment process</i>	59
<i>Participants</i>	60
<i>Measures</i>	60
<i>Procedure</i>	60
RESULTS.....	61
Paediatric Service Provision.....	61
Adult Service Provision.....	62
Thematic Analysis of Qualitative Responses.....	65
Theme 1. <i>Insufficient information and awareness about SRH in CF</i>	66
Theme 2. <i>Barriers to discussing SRH</i>	67
Theme 3. <i>Individual differences (inductive)</i>	68
Theme 4. <i>Roles and responsibilities of the CF team</i>	68
Theme 5. <i>Content</i>	69
Theme 6. <i>Age and time</i>	69
Theme 7. <i>SRH as part of CF overall (inductive)</i>	70
Theme 8. <i>The use of other sources of information</i>	71
DISCUSSION.....	71
Service Implications.....	73
Limitations.....	74
CONCLUSIONS AND FURTHER RESEARCH.....	74
REFERENCES.....	75
MAIN RESEARCH PAPER: The Relationship between Challenging Behaviour and Social Story Interventions: A Pilot Study in a Naturalistic Setting.....	78
LITERATURE REVIEW.....	79
Theories of Challenging Behaviour.....	79
Social Stories™.....	80
Evidence for Social Stories.....	80
Evidence for Factors Influencing Effectiveness.....	80
Treatment Response to Social Stories.....	82

Functional Assessment in Social Stories.....	83
Research Questions.....	84
Hypotheses.....	84
METHOD.....	85
Ethical Approval.....	85
Sample Size and Power.....	85
Participants and Recruitment.....	86
Measures.....	91
Procedure.....	91
Phase 1: Functional assessment.....	91
Phase 2: Baseline and intervention.....	92
Statistical Analysis/Design.....	93
RESULTS.....	97
Treatment Response and Function.....	97
Functional Congruence.....	97
Social Skills Questionnaire (SSQ).....	98
Social Story Structure.....	98
Correlations.....	98
Case Series Approach.....	99
Young Person Feedback Form.....	99
DISCUSSION.....	100
Strengths and Limitations.....	103
CONCLUSIONS.....	104
REFERENCES.....	106
EXECUTIVE SUMMARY: The Relationship between Challenging Behaviour and Social Story™ Interventions: A Pilot Study in a Naturalistic Setting.....	112
AIMS AND BACKGROUND.....	113
METHOD.....	113
RESULTS.....	114
CONCLUSIONS AND IMPLICATIONS FOR CLINICAL PRACTICE.....	115

CONNECTING NARRATIVE.....	116
MAIN RESEARCH PROJECT.....	117
Development.....	117
Recruitment and Process.....	118
Service User Involvement.....	118
Reflections and Contributions.....	119
SERVICE IMPROVEMENT PROJECT.....	119
Development.....	119
Ethics.....	120
Recruitment and Process.....	120
Service User Involvement.....	121
Reflections and Contributions.....	121
LITERATURE REVIEW.....	121
Development.....	121
Process.....	122
Reflections and Contributions.....	122
CASE STUDIES.....	123
CONCLUSIONS AND FUTURE RESEARCH.....	123
ACKNOWLEDGEMENTS.....	125
APPENDICES.....	127

List of Appendices

Appendix A	CRITICAL REVIEW OF THE LITERATURE.....	127
	Appendix A1. Codebook for Content Analysis.....	127
	Appendix A2. Submission Guidelines for the Journal of Clinical Child and Adolescent Psychology.....	134
Appendix B	SERVICE IMPROVEMENT PAPER.....	138
	Appendix B1. Relationships and Sexual and Reproductive Health in Cystic Fibrosis.....	138
	Appendix B2. Confirmation of Ethical Approval including Amendments by the University of Bath Psychology Ethics Committee.....	147
	Appendix B3. Ethical Approval from UHBristol Research and Development QIS Department.....	149
	Appendix B4. Ethics Documents for Participants.....	150
	Appendix B5. Journal Submission Guidelines for Sexuality and Disability.....	155
Appendix C	MAIN RESEARCH PAPER.....	161
	Appendix C1. Types of Sentences in a Social Story.....	161
	Appendix C2. Measures for the Social Stories Project.....	162
	Appendix C3. Case Series Summary Table of Statistics by Participant.....	170
	Appendix C4. Graphs of Treatment Response by Function.....	171
	Appendix C5. SPSS Table of Correlations between Proportion of Sentence Types and Magnitude (%) of Change in Frequency of Behaviour.....	173

Appendix C6. Ethical Approval from the University of Bath Psychology Ethics Committee.....	174
Appendix C7. Ethics Documents for Educational Staff.....	178
Appendix C8. Ethics Documents for Parents.....	183
Appendix C9. Ethics Documents for Young People.....	188
Appendix C10. Journal Submission Guidelines for Research in Developmental Disabilities.....	192

List of Tables

Critical Review of the Literature:	Table 1. <i>Key Assumptions and Cognitions in the Cognitive Model of SAD (Clark, 2001)</i>	22
	Table 2. <i>Results of Systematic Literature Search</i>	26
	Table 3. <i>Demographics and Methodological Data for Included Studies</i>	29
	Table 4. <i>Outcome of Content Analysis</i>	42
Service Improvement Paper:	Table 1. <i>Consultation Session Discussion Topics</i>	58
	Table 2. <i>Demographic Summary of Sample</i>	61
	Table 3. <i>Role of the Paediatric CF Service in SRE</i>	62
	Table 4. <i>Support around SRH</i>	62
	Table 5. <i>Information Requests to the Paediatric CF Service</i>	63
	Table 6. <i>Information Provision by the Paediatric CF Service</i> ...	63
	Table 7. <i>Topics of Information Sought from the Adult CF Service</i>	64
	Table 8. <i>Rationale for Not Seeking Information</i>	64
	Table 9. <i>Final Themes from Qualitative Data</i>	66
Main Research Paper:	Table 1. <i>Theoretical Underpinnings to Social Stories Proposed by Gray (2004)</i>	81
	Table 2. <i>Social Story Guidelines</i>	82
	Table 3. <i>Outcome of Meta-Analyses Reviewing Treatment Response to Social Stories</i>	83

Table 4. <i>Research Basis for Effect Size</i>	86
Table 5. <i>Demographic Characteristics of Sample</i>	87
Table 6. <i>Categories of Target Behaviours</i>	87
Table 7. <i>Screening Criteria for Study Participants</i>	88
Table 8. <i>Measures used in the Pilot Study</i>	91
Table 9. <i>Measurement of Study Variables</i>	93
Table 10. <i>Analysis Methods for Hypotheses Testing</i>	94
Table 11. <i>Treatment Response (TR) for Frequency, Duration and Severity of Behaviour</i>	96
Table 12. <i>Descriptive Statistics including Standardised Mean Difference (SMD) Effect Size within Treatment Response Categorisation</i>	96
Table 13. <i>2x3 Contingency Table for Function and Treatment Response</i>	97
Table 14. <i>2x2 Contingency Table for Functional Congruence and Treatment Response</i>	98
Table 15. <i>Outcome Pre and Post Intervention on the Social Skills Questionnaire (SSQ)</i>	98
Table 16. <i>2x2 Contingency Table for Violating Social Story Sentences and Treatment Response</i>	99
Table 17. <i>Comparison between Participants at the Extreme Ends of the Range of Treatment Responses</i>	100

List of Figures

Critical Review of the Literature:	<p><i>Figure 1.</i> Synonyms and relevant terms to illustrate the definition of cognitive factors. Based on Hofmann (2007) and created using Wordle (Feinberg, 2014)..... 21</p> <p><i>Figure 2.</i> PRISMA diagram outlining the process of literature selection. Adapted from: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). <i>Preferred Reporting Items for Systematic Reviews and Meta Analyses: The PRISMA Statement</i>. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097..... 28</p>
Service Improvement Paper:	<p><i>Figure 1.</i> (a) Prevalence rate of carriers of CF gene: 1 in 25 people. (b) Genetic map for two carriers of CF gene. Probability of child having CF: 1 in 4. (c) Genetic map of one individual with CF and one carrier of CF gene. Probability of child having CF: 1 in 2. Based on information from: Cystic Fibrosis Trust (2016c)..... 55</p> <p><i>Figure 2.</i> Participants' overall rating of provision by the CF service around SRH. No. of respondents = 31..... 65</p>
Main Research Paper:	<p><i>Figure 1.</i> Recruitment process for schools..... 89</p> <p><i>Figure 2.</i> Recruitment process for participants within each school setting..... 90</p> <p><i>Figure 3.</i> ABA+ study procedure including timescale and measures..... 92</p>

Abstracts

The Relationship between Challenging Behaviour and Social Story™ Interventions: A Pilot Study in a Naturalistic Setting

Background: Research into Social Stories™, an intervention to aid social understanding in children with Autism Spectrum Disorder (ASD), has shown variable effectiveness. Several factors have been proposed to influence treatment response, including function, functional congruence and adherence to social story guidelines.

Method: A case series ABA+ design was used. Participants were 16 young people aged 5-13 years with a diagnosis of ASD across 5 schools. Teachers completed questionnaires around behavioural function and social skills, and behaviour recording forms for 5 days before and after the social story intervention.

Results: Eleven participants showed reduced behavioural frequency, suggesting social stories were an effective intervention for challenging behaviour. Associations between treatment response and function and functional congruence were non-significant. Associations between other variables were also non-significant, including magnitude of change in frequency of behaviour, change in social skills, comprehension and social story structure.

Discussion: Findings challenges the influence of several factors previously associated with treatment response to a social story intervention. Considerations were also raised around feasibility of research in naturalistic settings, and the importance of adherence to social story guidelines was questioned. Areas for further research include investigating the independent contribution of social stories on challenging behaviour and social skills.

Keywords: Social Stories™; Autism Spectrum Disorder; challenging behaviour; social skills; functional analysis

Sex and Relationships Education for Individuals with Cystic Fibrosis: A Service-Directed Approach

Background: Increasing life expectancy within Cystic Fibrosis (CF) raises challenges around previously neglected topics such as sexual and reproductive health (SRH). The study aimed to gather retrospective experiences of service provision around SRH to consider the role of the CF service, age of information provision and unmet needs highlighting possible improvements to provision.

Method: *Phase 1:* An Adult CF team participated in a consultation session generating survey questions around SRH. *Phase 2:* A 22-item online survey was constructed and disseminated to adult CF patients.

Results: Unmet needs were found in SRH provision in paediatric and adult CF services, with further information required by patients on topics including parenthood and fertility.

Conclusions: Results support previous research findings highlighting the need for standardised provision around SRH. Age of SRH provision suggested individual differences in need within the paediatric service. Further research could explore format and specific age of SRH information provision.

Keywords: Cystic Fibrosis; sexual and reproductive health; sex and relationships education; paediatric; adult

Self-Report Cognitive Factors Maintaining a Failure to Speak in Selective Mutism

Background: Selective mutism, typically a childhood disorder, is characterised by a failure to speak in certain settings. Several theories exist around underlying mechanisms, including anxiety, behavioural inhibition and emotion regulation. Research into cognitive factors is limited; largely based on observer report. This review aims to address the gap in literature through synthesising evidence of cognitive factors based on participant self-report.

Method: A systematic literature search was conducted, using search terms and inclusion/exclusion criteria, with systematic extraction of demographic and methodological data. Self-report data relating to cognitive factors in a failure to speak was extracted and synthesised using qualitative inductive content analysis.

Results: Seventeen studies were included in the review. Seven main categories and seven sub-categories were identified, with a failure to speak due to anxiety being the most frequent cognitive maintenance factor. Other factors included beliefs about control of speech, the expectations of others about speaking and mutism as protective.

Discussion: Despite methodological issues and limited research, the review synthesises current evidence around cognitive factors in selective mutism, including factors beyond anxiety. Clinical implications include broader assessment and intervention planning around maintenance factors. Areas for further research include developing a cognitive model and constructing measures of cognitive processes in selective mutism.

Keywords: selective mutism; cognitive; self-report; maintenance; anxiety

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Self-Report Cognitive Factors Maintaining a Failure to Speak in Selective Mutism

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Intended journal: This paper is intended for publication in the Journal of Clinical Child and Adolescent Psychology. This journal publishes papers which contribute to the understanding of development and maintenance of clinical difficulties in young people.

This is therefore highly relevant for this paper which contributes knowledge around cognitive maintenance factors in selective mutism, a disorder predominantly of childhood.

Introduction

Description and History of Selective Mutism

Selective mutism (SM) is characterised in the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM 5, American Psychiatric Association (APA), 2013) as a “consistent failure to speak” (p.195) in certain social situations but not in others, which disrupts achievement at work or school, including social communication, and cannot be better explained by another factor such as comorbidity (including communication disorders) or limited knowledge of a language. Symptoms must be present for at least a month, excluding the first month of starting school (APA, 2013). Prevalence rates are estimated to fall between .47 and .76% (Viana, Beidel & Rabian, 2009) although estimates have varied according to the population studied, ranging from .11 – 2.2% (Hua & Major, 2016). Mean age of onset falls between two and five years, with symptoms typically manifesting in the school setting (Hua & Major, 2016; Muris & Ollendick, 2015). Research has suggested a mean duration of SM of eight years with naturalistic reduction in the primary symptom of a failure to speak over time (Muris & Ollendick, 2015), although research with adults is limited. Studies have also suggested a continuation of associated difficulties into adulthood even once the primary symptom has resolved, such as a higher incidence of communication problems, social difficulties and other psychiatric disorders, including social anxiety (Remschmidt, Poller, Herpertz-Dahlmann, Hennighausen, & Gutenbrunner, 2001; Steinhausen, Wachter, Laimböck, & Metzke, 2006).

Historically, SM was classified in the DSM-III (APA, 1980) under ‘Other disorders’, within disorders usually diagnosed in childhood (Newman, 2004). A shift in terminology took place in 1994 from “elective mutism”, suggesting a pervasive “refusal” to speak in DSM-III (APA, 1980); to “selective mutism” from DSM-IV (APA, 1994) onwards, recognising the presence of speech in some settings, and “failure” to speak not characterised by choice or wilfulness (Newman, 2004; Viana et al., 2009). The change in terminology led to a new understanding of SM, with research highlighting the importance of anxiety within the presentation, and this is reflected within the reclassification in the DSM 5 of SM as an anxiety disorder (APA, 2013).

Theoretical Understanding of Selective Mutism

Within the last decade, several reviews have explored and updated current conceptualisation of selective mutism, including theories of aetiology and maintenance which may underpin cognitive factors in SM from systemic, behavioural, psychodynamic, emotion regulation and developmental perspectives. A number of systematic reviews were published during the period of consultation around reclassification in the DSM 5 (APA, 2013), prior to its publication (e.g. Scott & Beidel, 2011; Sharkey & McNicholas, 2008; Viana et al., 2009; Wong, 2010). Two reviews have been identified since SM was reclassified as an anxiety disorder in DSM 5 (APA, 2013) which largely incorporated findings of the earlier papers. Hua and Major (2016) reviewed literature to provide an overview of the current understanding of selective mutism, including diagnosis, epidemiology, causes, prognosis and treatment. Maintenance factors were not explored. The review provided a comparison with social anxiety disorder (SAD or social phobia), highlighting evidence for similarities in symptomatology, as well as differences in developmental factors such as associations with language problems and bilingualism, and oppositionality. An earlier review by Muris and Ollendick (2015) provided an in-depth systematic review of literature associating SM with anxiety. The authors outlined research demonstrating high co-morbidity within samples between SM and SAD, as well as other anxiety disorders. In addition, the review highlighted elevated levels of anxiety amongst those with SM in control comparison studies, with similar levels reported to those with SAD without SM. However, the overlap in symptomatology raises challenges for researchers in identifying children with SM who do not also meet criteria for SAD in comparison studies.

The review also outlined the key aetiological theories in SM and evidence-base for each, including genetics, temperament (oppositionality and behavioural inhibition), environmental (home/school) and neurodevelopmental factors (Muris & Ollendick, 2015). This was conceptualised within a framework of anxiety leading to selective mutism, suggesting the influence of avoidance as a maintenance factor. As such, the authors concluded that whilst considerable evidence supports SM as an anxiety disorder, other potential factors not related to anxiety such as language difficulties and oppositionality should also be considered. In addition, the review explored evidence on both sides of the debate around whether SM should be considered an extreme form of social anxiety, or a

distinct anxiety disorder relating to a specific phobia of speaking (for example Omdal & Galloway, 2008 who found that children were still able to engage in social situations using nonverbal communication, contrasting with a general fear of negative social evaluation in social anxiety disorder).

Other important contributions from earlier reviews include Scott and Beidel's hypothesis (2011) around emotion regulation theory as a framework to understand failure to speak. The authors suggested that differences in emotion regulation following distress may explain why failure to speak develops as a functional emotion regulation strategy (or avoidance behaviour) in a small number of children, but not all who are socially anxious. The authors contrast the use of an active coping strategy with explanations based on an inability to speak as a consequence of high anxiety.

Beyond anxiety, other aetiological theories for SM were synthesised by Wong et al. (2010). Key concepts within each explanation included selective mutism as a result of unresolved conflicts in childhood (psychodynamic theories); a manifestation of behavioural inhibition due to activation of the sympathetic nervous system, or a learned adaptive response to the environment (behavioural theories); failure to speak as a result of interdependent attachment relationships (family systems theory) and mutism as a response to trauma (see Wong et al., 2010 for details). A developmental psychopathological explanation was also hypothesised involving the interaction of environmental factors (such as those highlighted in other theories) with an anxious predisposition (Wong et al., 2010). Minimal research has been conducted into maintenance factors within SM beyond a failure to speak as an avoidance behaviour (Muris & Ollendick, 2015).

Intervention for Selective Mutism

Reviews of treatment effectiveness for SM have generally supported the use of behavioural and cognitive behavioural therapy, with some support for pharmacological approaches (Cohan, Chavira & Stein, 2006; Hua & Major, 2016; Muris & Hollendick, 2015; Wong et al., 2010). Hua and Major (2016) found the strongest evidence-base for the use of CBT in SM, based on a review by Cohan, Chavira and Stein (2006), although the majority of interventions were behavioural in nature and consisted mostly of case reports. At present, a cognitive model has not been developed for SM, therefore cognitive behaviour therapy (CBT) treatment protocols for SM are adapted based on a general

cognitive model of anxiety, or specific adult models of SAD (Clark, 2001), raising questions about consistency of interventions across studies.

In addition, cognitive models of SAD were not applied to young people until recently, with previous approaches proposing an underlying social skills deficit as maintaining social anxiety (Cartwright-Hatton, Hodges, & Porter, 2003). Research has begun to investigate the application of a cognitive model to SAD in young people, for example demonstrating the presence of negative beliefs about performance amongst young people (Cartwright-Hatton et al., 2003; Cartwright-Hatton, Tschernitz, & Gomersall, 2005). However, further work is needed to adapt the cognitive model; highlighting further questions about the applicability of CBT based on adult models of SAD to a disorder predominantly found in childhood (SM). In addition, application of CBT underpinned by a cognitive model of SAD assumes that the primary symptom of selective mutism (a failure to speak) is maintained by the same cognitive factors (see Table 1). Systematic searching of the literature suggests that specific cognitive factors in selective mutism have not yet been reviewed, highlighting a gap in the literature.

Furthermore, considerable methodological challenges exist with collecting data directly from participants with SM who may not communicate with researchers. As such, much of the theoretical evidence is based on observer reports from parents or teachers of those with SM, for example through the use of the Child Behaviour Checklist (Achenbach & Edelbrock, 1983) or the Selective Mutism Questionnaire (Bergman, Keller, Piacentini, & Bergman, 2008). However, the validity of making inferences about thought processes based on observable behaviour alone, rather than experiential evidence, is questionable. An observed behaviour such as not speaking may score on a measure of shyness, as well as social isolation/exclusion, despite the function (or cognitions) behind the failure to speak remaining unknown (Walker & Tobbell, 2015). Therefore, it is hypothesised that in order to gain an accurate insight into cognitive factors in SM, self-report evidence is required from individuals with SM who are able to report on their direct experience of factors maintaining a failure to speak.

Rationale

Studies have begun to make use of self-report methodology, however extensive searching of the literature suggests that self-report evidence from those with selective

mutism has not yet been reviewed. This review therefore aims to address two gaps in the literature by examining evidence for cognitive factors underlying a failure to speak in SM, based on participant's own experiences gathered through self-report. For this review, cognitive factors were operationalised as: mental processes or actions which impact on and reinforce the behaviour of not speaking ("Cognition," n.d.; Hofmann, 2007). Specifically, mental processes refer to an individual's perception of self, world and others. Synonyms and other relevant terms have been illustrated in Figure 1. This evidence will be considered in light of current conceptualisations and theories of SM, for example relating to SAD. Implications for practice and recommendations for further research will be considered.



Figure 1. Synonyms and relevant terms to illustrate the definition of cognitive factors. Based on Hofmann (2007) and created using Wordle (Feinberg, 2014).

Method

Review Design

A systematic approach was taken to literature searching using strictly defined search terms and inclusion/exclusion criteria, based on the working definition of cognitive factors. Systematic data extraction was also employed around demographics and methodology. A qualitative approach to synthesising the literature was taken due to a lack of research studies or quantitative measures directly examining cognitive factors in SM. A small number of qualitative studies have been conducted into subjective experiences of those with SM (largely retrospectively with adults), and the subjective experience of

participants has been captured in case reports where this has not been the primary focus of the research. As such, inductive qualitative content analysis was identified as a suitable method of synthesis in order to categorise and quantify self-reported cognitive factors across studies (Dixon-Woods, Agarwal, Jones, Young, & Sutton, 2005; Elo & Kyngas, 2008; Finfgeld-Connett, 2013; Graneheim & Lundman, 2004). The process of content analysis involved defining units of analysis, data extraction, coding of extracts and categorisation. The author was aware of the influence of their own contextualist epistemology in the content analysis. Shaped through experience and training from a cognitive standpoint within a scientist-practitioner framework, data extraction and categorisation was influenced by a commitment to developing evidence-based knowledge through capturing an individual's lived experience rather than researcher interpretation.

Table 1

Key Assumptions and Cognitions in the Cognitive Model of SAD (Clark, 2001)

Cognitive model of Social Anxiety Disorder (SAD)	
Assumptions	Consequences of assumptions
Excessively high standards for social performance	Appraisal of social situations as dangerous
Conditional beliefs concerning the consequences of performing in a certain way	Predictions of failing to reach desired level of performance
	Interpretation of neutral cues as signs of negative evaluation by others
Unconditional negative beliefs about the self	Evaluation of self as social object through self-monitoring and internal focus of attention, used to make judgements about how they appear to others, maintained by safety behaviours

Inclusion Criteria

Participants.

Inclusion criteria within each study sample were participants of any age (including adults) and any gender and ethnicity with a current or past primary diagnosis of selective mutism, as insight into cognition requires direct report from those experiencing SM. Although a minimum age was not specified, the requirement for self-report may have acted as a naturalistic filter. A maximum age cut-off was not specified to enable the use of retrospective reporting from adults, as it is anticipated that adults may be more able to identify cognitive factors.

Intervention and outcome.

Intervention for selective mutism was not a requirement for the review, as the research question examined cognitive factors; evidence for which may be obtained through assessment/research interview in the absence of an intervention. Inclusion criteria for outcome was any mode of self-report relevant to cognitive factors in a failure to speak by participants with a current or past diagnosis of selective mutism, such as interviews, quotes and lists of feared situations involving speaking. Self-report minimises possible bias from inferences made by observers about the internal experiences (cognitions) of those with SM.

Study design.

All participant designs were included (e.g. case studies, group designs) due to the rarity of selective mutism and the paucity of well-controlled and adequately powered studies. In addition, qualitative designs were hypothesised to be more suitable for gathering evidence relating to participant experience. Similarities in findings across several single-case studies may provide evidence for cognitive factors or provide additional support for findings from larger group studies.

Exclusion Criteria

Search parameters were limited to studies published from 1994 to 2017. This reflected the year of reclassification of elective mutism as selective mutism in DSM-IV (APA, 1994). Prior to 1994, classification of elective mutism was not associated with anxiety, and was defined as a refusal to speak, which conflicts with current understanding

of selective mutism as a ‘failure to speak’ (DSM 5; APA, 2013), categorised as an anxiety disorder.

Participants.

Participants where selective mutism was classified within the study as a manifestation of another disorder (for example Post-Traumatic Stress Disorder), or those with a co-morbid developmental disorder (e.g. Autism Spectrum Disorder, learning disability, genetic disorders) were excluded, as comorbid disorders may be a moderator of cognition over and above the influence of selective mutism.

Intervention and outcome.

Studies were excluded when only behavioural report was available, for example number of utterances made by the participant, as this does not give insight into cognitive factors therefore is not relevant to the question. Studies were also excluded when self-report was not specific to a failure to speak, for example the inclusion of a diagnostic or anxiety measure without providing examples relating to speaking.

Study design.

Non-research articles were excluded (such as systematic reviews) due to a reliance on self-report participant data for the review question. Unpublished (grey) literature was also excluded due to concerns about the validity of findings from research that has not been subject to peer review.

Information Sources

The following databases were searched within the publication date parameters of 1994 to January 2017:

1. Pubmed
2. Psycnet
3. Web of Science

Non-English papers were excluded. Identical search terms and processes were used to search each database and records kept of the number of studies extracted (Table 2).

Search terms were based on Hofmann's paper (2007) as shown in Figure 1. The sequence of search terms was as follows:

1. Publication dates were set as 1994 to 2017.
2. Initial search terms were entered on a single line as: "selective mutism" OR "elective mutism" with the criteria of 'All fields'. A record was made of the number of returned entries (Table 2).
3. In addition to the initial search terms, the process was repeated on a second line with a record made of returned entries with the addition of each pair of search terms. All results were exported to Endnote online. The search was constructed as follows: (All fields) "selective mutism" OR "elective mutism":
 - a. AND (All fields): "cognitive" OR "cognition"
 - b. "cognitive" OR "cognition" OR "belief" OR "beliefs"
 - c. "cognitive" OR "cognition" OR "belief" OR "beliefs" OR "thought" OR "thoughts"
 - d. "cognitive" OR "cognition" OR "belief" OR "beliefs" OR "thought" OR "thoughts" OR "maintenance" OR "maintaining"
 - e. "cognitive" OR "cognition" OR "belief" OR "beliefs" OR "thought" OR "thoughts" OR "maintenance" OR "maintaining" OR "attribution" OR "attributions"
 - f. "cognitive" OR "cognition" OR "belief" OR "beliefs" OR "thought" OR "thoughts" OR "maintenance" OR "maintaining" OR "attribution" OR "attributions" OR "perception" OR "perceptions"

Additional studies were identified through handsearching the reference lists of review papers excluded during the screening process which were relevant to the subject area. Reference lists were searched using the same systematic process of inclusion/exclusion criteria as the initial screening process. One additional study was identified through researching background literature for the review and screened using the same process.

Data Collection and Analysis

Study Selection.

All retrieved titles and abstracts were assessed for relevance to the research question (on the topic of selective mutism with scope for exploration of cognitive factors, including reference to anxiety). All abstracts and titles were subsequently screened according to the inclusion/exclusion criteria. If insufficient information was gathered from the abstract, papers were accessed for full-screening and assessed according to the inclusion/exclusion criteria (see Figure 2 for a flow chart of included/excluded studies). A second independent rater assessed papers highlighted for full-text screening (excluding those identified through other methods; n=46) obtaining inter-rater reliability of 98% with discrepancies resolved through discussion.

Table 2

Results of Systematic Literature Search

Search terms (publication dates: 1994-2017)		Database			Total exported to Endnote (PsycExtra papers not exported to Endnote)
Line 1 (all fields)	AND Line 2 (all fields)	Pubmed	Web of science	Psycnet	
"selective mutism" OR "elective mutism"		193	308	445	
"	cognitive OR cognition	36	45	74	
"	"OR belief OR beliefs	39	49	77	
"	"OR thought OR thoughts	46	54	98	
"	"OR maintenance OR maintaining	48	65	108	
"	"OR attribution OR attributions	48	65	109	
"	"OR perception OR perceptions	54	66	119	233 (6)
				Minus duplicates:	159 (6)

Data Extraction Process

A data extraction table was created using Excel (guided by Noyes & Lewin, 2011) in order to extract methodological data, for example relating to the sample, data collection approach and data analysis (see Table 3). A subsequent data extraction table was developed in Excel to extract qualitative data specific to the research question through the process of inductive content analysis (Table 4).

All selected studies were read and re-read several times to gain an overview of the topics and findings of the research. Relevant data was identified using the following rules for extraction: first-order data only (i.e. self-report or researcher report of participant's response, not researcher interpretation); extracts contained a cognitive factor according to the working definition; extracts directly referenced speaking within the unit of analysis or immediate context of the surrounding paragraph. This was to ensure data related to cognitive factors around a failure to speak, rather than general cognitive factors for example relating to social situations. The unit of analysis was defined as a mention of a cognitive factor, including necessary context. This typically took the form of a sentence when reported by a researcher, a complete hierarchy when using a written structured response or a block quotation for direct self-report. Block quotations were only divided when more than one cognitive factor was identified within the extract.

All data extracts were copied into a codebook in Excel. The software program NVivo 11 (2015) was also used to support the data extraction process. The researcher assigned a code to each data extract which summarised the cognitive factor in the extract. As coding continued, sub-categories to cluster codes were identified and added to the codebook. Once all extracts were coded, the researcher clustered codes into the sub-categories, and checked for any codes that could not be appropriately assigned. The researcher then identified any over-arching main categories which united more than one sub-category. In line with an inductive process of identifying categories for content analysis, the process was fluid, involving moving back and forward between the original studies, codes and categories in order to generate categories which reliably captured all the data. Main categories were validated by a second researcher who assigned data extracts (n=102) to categories with an inter-rater reliability level of 79%. Any discrepancies were discussed until agreement was reached.

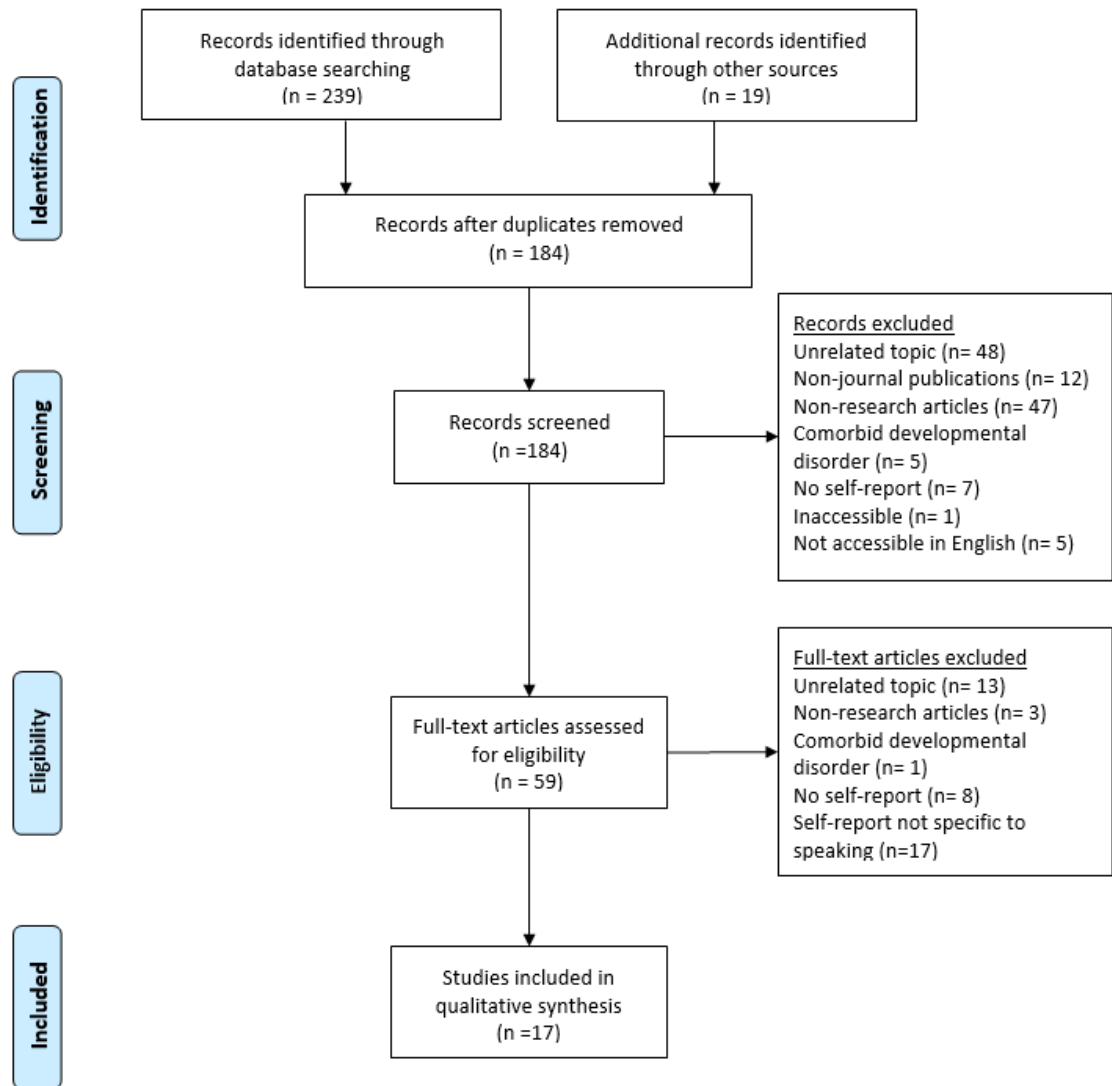


Figure 2. PRISMA diagram outlining the process of literature selection. Adapted from: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *Preferred Reporting Items for Systematic Reviews and Meta Analyses: The PRISMA Statement*. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

Table 3

Demographics and Methodological Data for Included Studies

No.	Reference	Purpose of study	Sample characteristics				Sampling approach	Study design	Data collection and analysis for relevant data
			Sample size	Age, gender, background	Diagnostic criteria and co-morbidities	Sample setting			
1	Albrigtsen, Eskeland & Mæhle, 2016	Opportunity to explore lived experience of SM, including understanding of how it developed, its impact on social and family life, and the recovery process.	2	Male, MZ twins, adolescent	School psychology services: neuropsychological assessment, clinical observations and diagnostic tests (not specified) to confirm diagnosis of SM. Comorbid specific language disorder.	Norway. Treatment in inpatient family unit. Follow-up interview at family home.	Opportunistic - approached treatment team following therapy.	Qualitative	Semi-structured interview with four main themes and follow-up questions. Chronological interview from time of stopping talking, ending with time of interview. Thematic analysis.
2	Baptiste, 1995	To describe psychotherapeutic treatment of a lesbian "stepfamily" with a 9 yr old child who presented with EM	1	Female, 9yrs, "lesbian stepfamily"	No formal diagnosis described or included as part of treatment; no comorbidities reported.	Self-referred, no description of therapy setting. USA	Self-referred, selection process not described	Case report	Clinical interview throughout sessions
3	Boon, 1994	To support view in research that SM should be considered an anxiety spectrum disorder with a case study	1	Female, 6 yrs, Caucasian	Not reported	US; study setting not given	Not reported	Case report	Clinical interview

4	Christon, Robinson, Arnold, Lund, Vrana & Southam, 2012.	Present the treatment of a case of SM in a female adolescent using an evidence-based CBT modular treatment for childhood anxiety disorders (MATCH).	1	Female, 15, Latina American	Met DSM-IV-TR criteria for SM using Kiddie Schedule for Affective Disorders and Schizophrenia-Present and Lifetime version. Additional info from Selective Mutism Questionnaire, Child Behaviour Checklist, Youth Self Report, Achenbach system of empirically based assessment. Comorbid social phobia (DSM-IV-TR)	USA. University-based clinic for treatment of youths and adults with anxiety disorders	Referral to clinic - selection criteria not reported	Case report	Diagnostic interview: clinical interview (assessment) using written responses, Subjective Units of Distress (SUDS); fear hierarchy
5	Giddan, Ross, Sechler & Becker, 1997	To report on efficacy of treatment to target response initiation	1	Female, 8 yrs, Father spoke Spanish in the home - left when child was 2yrs old	DSM-IV criteria provided but no report of how symptoms were compared to this. Speech and academic delay reported - no formal diagnosis.	Public school special education program for children with severe behaviour handicaps within a medical school psychiatry department	Not reported	Case report	Clinical interview

6	Jackson, Allen, Boothe, Nava, & Coates 2005	To describe a brief multi-dimensional approach to treating SM.	1	Male, 6 yrs	No description of use of formal criteria to confirm diagnosis of SM. No other formal diagnoses.	US - psychological services.	Referral to clinic - selection criteria not reported	Case report	Feelings thermometer. Fear hierarchy through client shaking/nodding head in response to list of symptoms - 'why was he here'?
7	Kehle, Madaus, Baratta & Bray, 1998	To report on efficacy of a packaged procedure of treatment for SM using AB design.	2 (one excluded due to 'cognitive disability')	Female, 9yrs, Caucasian; male, 5 yrs, Caucasian	DSM-IV criteria for SM. IQ provided to rule out developmental disorder. Comorbid diurnal enuresis.	US - unknown setting	Not reported	Series of single case baseline-intervention (AB) design, assessment of quality according to comparison with experimental designs	Clinical interview end of therapy and follow-up - asked why they did not talk

8	Omdal, 2007	To explore the child's experience of selective mutism and the recovery process, and consider implications for diagnosis and treatment.	6	Female, adults who were selectively mute in childhood, Norwegian, one participant grew up in South Africa, another born in France; included MZ twins.	Assessed as part of study to meet DSM-IV criteria for SM based on 'information offered'. Comorbidities: Social anxiety disorder (SAD); two, self-reported); depression (one self-report), suicidal behaviour (one), acute self-consciousness (one).	Norway. Participant home or chosen familiar location	Opportunity sample following interview with researcher on daytime radio inviting contact by participants who had experienced SM, or who had a child with SM	Qualitative	Semi-structured interview about retrospective experiences - full interview schedule included with the paper. Thematic analysis.
9	Omdal & Galloway, 2008	To consider whether SM can be conceptualised as social phobia or specific phobia; to provide a theoretically derived basis for intervention; to consider whether progress of children in study is consistent with theoretically derived predictions.	6 adults with SM; parents of 5 children with SM (no self-report)	Female, adults who were selectively mute in childhood	Had met DSM-IV criteria for selective mutism as children and adolescents. No comorbidities reported.	Norway.	Not reported	Qualitative	Semi-structured interview about retrospective experiences - full interview schedule included with the paper. Thematic analysis

10	Remschmidt, Poller, Herpertz-Dahlmann, Hennigause & Gutenbunner, 2001	To conduct a follow-up study of patients with SM to extend data on course of SM and predictors of outcome and provide a more extensive description of symptoms specific to mutism patients.	45 initial patients; 41 follow-up; 31 investigated personally; 25 with subjective experiences (included in the review)	Male (23); Female (22); at time of referral: 8.7 yrs +/- 3.6 yrs. Follow-up = average age = 20.5yrs +/- 6.7yrs. Social class: 31 = lower social class; 10 = middle class; 2 upper class; 2 unclassified	Reclassified from original patient records using DSM-III-R criteria. Comorbidities: Marburg Symptom checklist at follow-up. Reported comorbidities e.g. anxiety, depression	Germany. Department of Child and Adolescent Psychiatry and child guidance clinic	All patients referred between 1964 and 1979 - 4 excluded due to not meeting criteria when reclassified e.g. learning disability	Matched clinical control comparison design	Mannheim Biographic Inventory + The Biographic Inventory. Standardised interview about subjective experiences for 25 patients at follow-up
11	Reuther, Davis, Moree & Matson, 2011	To demonstrate a modular CBT approach to selective mutism.	1	Male, 8 yrs, Caucasian	Anxiety Disorders Interview Schedule-IV (ADIS-IV) parent interview - met DSM-IV criteria for SM. Comorbid social phobia (DSM-IV via ADIS-IV).	US - doesn't state setting	Referral to clinic - selection criteria not reported	Case report	Fear hierarchy with ratings of anxiety from 0-10

12	Rye & Ullman, 1999	To describe the treatment of an adolescent with long-term mutism. To provide some potentially useful ways to conceptualise and treat long-term intractable cases of SM.	1	Male, 13 yrs	Not reported	USA - University-based psychology clinic	Referred to university-based clinic - selection criteria not reported	Case report	Clinical interview. Hierarchy of feared events rated using SUDS
13	Vecchio & Kearney, 2007	To describe and discuss a case of SM in a young Hispanic female whose parents spoke only limited English.	1	Female, 10 yrs, Hispanic, parents spoke limited English	Structured diagnostic interview ADIS-IV. Bilingual. Generalised anxiety disorder (GAD); SAD. Subclinical symptoms of specific phobia, separation anxiety disorder.	USA - Specialist university-based clinic for anxiety disorders	Referred by parents and school officials - selection criteria not reported	Case report	ADIS-IV diagnostic interview, nodding yes/no

14	Vecchio & Kearney, 2009	To examine the specific impact of exposure-based practice and contingency management in youths with SM.	9	Female (7), Male (2); 4-9 yrs; European-American (4); biracial; Asian-American (2); Hispanic (1)	All children met diagnostic criteria for SM using ADIS-IV - parent report and child report over 9yrs. Comorbidities: Social phobia (9), separation anxiety, specific phobia, Attention Deficit Hyperactivity Disorder (ADHD), enuresis, GAD and Oppositional defiance disorder (ODD).	USA, Clinic and university based assessments	Recruitment through schools, preschool and general press release to the community	Single-participant based alternating treatments design (case report)	Clinical interview at follow-up.
15	Walker and Tobbell, 2015	To gain an in-depth insight into SM from those who experience the difficulty and in doing so to further enrich understanding through an examination of the lived phenomenon in all its complexity.	4 + 1 (author)	Male (3), Female (2), 21-30 yrs	Had been diagnosed with SM (doesn't specify). Comorbid depression.	Participant homes - online study. Recruited through online SM communities. UK.	Self-selected through online communities. Screened for SM diagnosis, still experiencing difficulties, not in current treatment, able to communicate using non-spoken methods.	Qualitative	2x1 hour Semi-structured interviews using Skype instant messaging. Interpretive Phenomenological analysis

16	Wright, Cucarro, Leonhardt, Kendall & Anderson, 1995	To describe the treatment of a preschool girl with selective mutism and her family via a multimodal approach that includes family therapy, behavioural therapy, play therapy, and pharmacotherapy.	1	Female, 4 yrs 10 months, Caucasian	Not reported	US; outpatient preschool diagnostic and intervention day program	Referral to clinic - selection criteria not reported	Case report	Clinical interview
17	Yanof, 1996	Not stated	1	Male, 4.5yrs, "well-educated parents"	Not reported	Not stated	Not reported	Case report	Clinical interview/analysis

Note. Abbreviations used in table: SM = selective mutism; MZ = monozygotic; EM = elective mutism; USA = United States of America; US = United States; CBT = cognitive behaviour therapy; MATCH = Modular Approach to Therapy for Children With Anxiety, Depression, Trauma, or Conduct Problems; DSM-IV-TR = Diagnostic and Statistic Manual 4th edition text revision; DSM-IV = Diagnostic and Statistic Manual 4th edition; AB = baseline, intervention; IQ = intelligence quotient; DSM III-R = Diagnostic and Statistic Manual 3rd edition revised; UK = United Kingdom

Results

Quality of Studies

A quality appraisal tool was not used as evidence is mixed as to the usefulness of this process with qualitative data (see Dixon-Woods, Shaw, Agarwal, & Smith, 2004). In addition, the process of content analysis involved drawing conclusions based on quantified frequency of categories across studies, rather than weighting conclusions based on quality. However, quality appraisal was considered throughout the process of content analysis. Methodological approaches to data gathering were varied, from formal methods such as qualitative semi-structured interviews, to data gathered through standardised assessments and written idiographic measures (such as fear hierarchies), to informal clinician reports of data gathered during clinical interviews. As such, the majority of studies lacked methodological rigour, therefore limiting conclusions that can be drawn from extracted data.

In addition, for the majority of studies, extracted data consisted of raw qualitative data that had not been analysed as part of the results, for example a brief quote at follow-up about the child's thoughts on why they had been unable to talk. In order to increase consistency and comparability, a standardised process was therefore applied across all studies to quantify and categorise raw self-report data relevant to cognitive factors, rather than researcher interpretations or themes based on the data (where reported). This approach aimed to reduce bias of interpretations based on different theoretical stances across studies (for example psychodynamic compared to systemic), although epistemology may have had an impact on the information that researchers chose to report.

Demographic and Methodological Issues

Out of a total of 17 studies; ten studies employed a case study design; two studies used single case experimental design methodology including one case series; four studies employed a qualitative design using semi-structured interviews; and one study employed a matched clinical control comparison design.

Characteristics of Sample

Recruitment largely took place through referral to a clinic (nine studies), although selection criteria following referral was not reported for seven of these studies, and five

further studies did not report a recruitment strategy. Two studies used media advertising to recruit a sample, with additional recruitment used by one of these studies in schools and preschools. The remaining study used self-referral through online communities and selected participants who fulfilled screening criteria. Total sample size of participants where data was included in the review was 65; ranging from 1 to 45 participants across studies (with 25 of the largest sample providing data relevant to the review question).

Characteristics of Participants

Participant age ranged from 4 years old to adults (ages not reported), with thirteen studies using participants under 18, and four studies using adult participants. Demographic information about cultural or family background was reported in twelve studies, including information about 'race', class, culture (including parental), sexual orientation, family structure and parental education (see Table 3). A formal diagnosis of SM according to DSM criteria was reported in eight studies, with one additional study using formal diagnostic tests without specifying diagnostic criteria. The remaining studies did not specify how a diagnosis of SM was made.

Content Analysis

Qualitative inductive content analysis was used to develop categories by which extracted data was organised and quantified (Appendix A). The process resulted in seven main categories, within which were seven sub-categories (see Table 4). The occurrence of each category and the number of extracts within each category was counted across studies and summarised in Table 4. There were a total of 17 studies included in the analysis, of which:

- Four studies reported direct quotations from semi-structured interviews (face-to-face or online)
- Two studies reported specific responses by participants within a standardised assessment (e.g. diagnostic interview/questionnaire)
- Two studies reported idiographic measures of anxiety constructed by the participant (e.g. fear hierarchy)
- Seven studies reported cognitive factors cited by participants using informal methods during sessions (clinical interview, case report)

- Two studies reported participant responses using multiple methods (standardised assessment, fear hierarchy, clinical interview)

Categories for content analysis.

Failure to speak due to anxiety.

Across all studies, the most frequently cited cognitive factors related to anxiety, in 12 studies (71%). Cognitive factors maintaining a failure to speak fell into three categories: fears specific to speaking, fear of evaluation and other fears.

Fear of interactions involving speaking were identified in seven studies (41%). Data in this category was extracted from five studies in the form of standardised assessments or written fear hierarchies, with anxiety ratings provided for specific situations involving speaking (Christon et al., 2012; Jackson, Allen, Boothe, Nava, & Coates, 2005; Remschmidt et al., 2001; Reuther, Davis, Moree, & Matson, 2011; Rye & Ullman, 1999). Direct quotes from participants suggested being “afraid to talk” (Wright, Cuccaro, Leonhardt, Kendall, & Anderson, 1995), and identified the “stress and anxiety” and “uncomfortable” feelings associated with speaking (Walker & Tobbell, 2015).

Fear of evaluation was identified in six studies (35%). Two studies identified direct experiences of negative evaluation associated with current failure to speak (Albrigtsen, Eskeland, & Maehle, 2016; Omdal, 2007), whereas five studies reported fear of evaluation, including being “self-conscious” and not wanting others to “focus attention” on them or notice changes in communication (Omdal, 2007; Omdal & Galloway, 2008) and fear of negative social consequences (Rye & Ullman, 1999; Walker & Tobbell, 2015).

Other fears relating to a failure to speak were identified in six studies (35%), with shyness identified in three studies (Kehle, Madaus, Baratta, & Bray, 1998; Rye & Ullman, 1999; Wright et al., 1995). Other fears related to performance, for example getting things wrong (Christon et al., 2012), forgetting how to talk to others (Walker & Tobbell, 2015) and a specific fear of teachers (Omdal, 2007).

Beliefs about control over speaking.

Beliefs about control over speaking were mentioned in nine studies (53%) in the review. Within this, beliefs about lacking control represented the largest sub-category,

identified in seven studies (41%). Participants reported the perception of being unable to speak, as if there were an external force preventing them; referred to in a number of ways, such as a “mountain” (Omdal, 2007); “my brain” (Boon, 1994); a “subconscious roadblock”; “something in my head” and “selective mutism” (Walker & Tobbell, 2015), in contrast with their desire to speak. Failing to speak was therefore perceived as something “uncontrollable” (Christon et al., 2012), rather than a choice. In three studies, the experience of twins with selective mutism was reported, with beliefs around “locking” one another into silence, suggesting speech was out of the control of one twin without a response by the other twin (Albrigtsen et al., 2016; Omdal, 2007; Omdal & Galloway, 2008).

By contrast in five studies (29%), beliefs about having conscious control over speech were identified. Social and environmental factors were cited as reinforcing a decision not to speak (for example the perception that choosing to speak would allow others to “win”; Omdal, 2007; and describing participants as “determined” in their “refusal” to speak when under pressure to do so; Omdal & Galloway, 2008). One study highlighted a failure to speak as a functional response to the situation (“I want to go to work with Mummy”; Wright et al., 1995).

Beliefs about mutism as protective.

The category of mutism as protective was identified in six studies (35%). Extracts suggesting mutism as self-protective were identified in four studies (24%) and protective for others in two studies (12%).

Within self-protection, general beliefs around learning to stay silent and following rules (Giddan et al., 1997; Omdal, 2007) were identified, as well as specific beliefs around the function of mutism in protecting them from others and negative experiences (Omdal, 2007; Remschmidt et al., 2001). One study also identified the positive consequences of mutism such as “acceptance and friendliness” in contrast with negative consequences of speaking (Albrigtsen et al., 2016).

Two studies reported cognitive factors about protecting others by not speaking, for example the risk of revealing a family secret (Baptiste, 1995) or the power of speaking to bring harm to others, which was interpreted by Yanof (1996) based on the context of a

child's self-report ("I have a magic mouth") although clarification of the meaning was not provided by the child, therefore categorisation may not be reliable.

Negative self-perception.

Negative perceptions of self were identified in five studies (29%), including association of mutism with self-judgement through shame, humiliation and self-blame (Albrigtsen et al., 2016; Christon et al., 2012; Remschmidt et al., 2001; Walker & Tobbell, 2015), as well as being different from others (e.g. "my voice sounds strange"; Boon, 1994; and recognising experiences with mutism as "not normal"; Walker & Tobbell, 2015).

Expectations of others.

Beliefs about the expectations of others maintaining a failure to speak were identified in four studies (24%).

Beliefs around others not expecting participants to talk were identified in three studies (Albrigtsen et al., 2016; Omdal, 2007; Walker & Tobbell, 2015). This included a conditional assumption by participants themselves in one study (Albrigtsen, 2016) around being able to engage with social interactions only when they are not expected to respond.

By contrast, two studies identified participant beliefs that others would expect them to talk (Omdal, 2007; Omdal & Galloway, 2008), which meant they "withdrew more and more" due to anxiety about talking and being "unable to talk" (Omdal, 2007), and maintaining predictions that communication with others is "difficult" (Omdal & Galloway, 2008).

Perception of separate identity as mute.

Perceptions of mutism as a separate identity was identified in two studies (12%). Extracts included perceived roles by others which participants fitted into (e.g. "silent girl") or believed to be normal (Omdal, 2007), as well as participants perceiving their own identity as defined by mutism and seeking to retain this ("the girls who did not speak") so that others would not "win" (Omdal, 2007; Omdal & Galloway, 2008). By contrast, one study described the perception of being "different" as a result of mutism, but not "on the outside of the group" despite the difference (Omdal & Galloway, 2008).

Beliefs around the reason for mutism being unknown.

Across two studies (12%), the reason for mutism was identified as unknown, described in Remschmidt et al.'s study (2001) as "strange and unexplainable".

Table 4

Outcome of Content Analysis

Main categories (sub-categories)	No. of studies	% of studies	No. of mentions	References
Failure to speak due to anxiety	12	70.59%	32	1,4,6,7,8,9,10,11,12,13,15,16
<i>Fear of interactions involving speaking</i>	7	41.18%	16	4,6,10,11,12,15, 16
<i>Fear of evaluation</i>	6	35.29%	8	1,8,9,12,13,15
<i>Other fears</i>	6	35.29%	8	4,7,8,12,15,16
Control over speech	9	52.94%	28	1,3,4,8,9,14, 15, 16, 17
<i>Lack of control</i>	7	41.18%	20	1,3,4,8,9, 15, 17
<i>Active control</i>	5	29.41%	8	4, 8,9,14,16
Beliefs about mutism as protective	6	35.29%	16	1,2,5,8,10,17
<i>To self</i>	4	23.53%	11	1,5,8,10
<i>To others</i>	2	11.76%	5	2,17
Negative self-perception	5	29.41%	6	1,3,4,10,15
Expectations of others	4	23.53%	10	1,8,9,15
Perception of separate identity as mute	2	11.76%	7	8,9
Beliefs around the reason for mutism being unknown	2	11.76%	3	7,10

Note. Numbered references: 1. Albrigtsen, Eskeland & Maehle, 2016. 2. Baptiste, 1995. 3. Boon, 1994. 4. Christon, Robinson, Arnold, Lund, Vrana & Southam, 2012. 5. Giddan, Ross, Sechler & Becker, 1997. 6. Jackson, Allen, Boothe, Nava, & Coates 2005. 7. Kehle, Madaus, Baratta & Bray, 1998. 8. Omdal, 2007. 9. Omdal & Galloway, 2008. 10. Remschmidt, Poller, Hertz-Dahlmann, Hennigause & Gutenbunner, 2001. 11. Reuther, Davis, Moree & Matson, 2011. 12. Rye & Ullman, 1999. 13. Vecchio & Kearney, 2007. 14. Vecchio & Kearney, 2009. 15. Walker and Tobbell, 2015. 16. Wright, Cucarro, Leonhardt, Kendall & Anderson, 1995. 17. Yanof, 1996.

Discussion

In summary, according to participant self-report across 17 studies, the most frequently identified cognitive factors maintaining a failure to speak related to anxiety (12 studies; 71%), particularly beliefs about speaking as anxiety-provoking (seven studies; 41%); and beliefs about control over speech (nine studies; 53%), specifically the belief of lacking control over the ability to speak (seven studies; 41%). Other cognitive factors were categorised as beliefs about the expectations of others, perception of mutism as a separate identity, beliefs about mutism as protective and negative self-perception. An understanding of mutism was reported as unknown in two studies (12%).

Despite evidence suggesting promising treatment outcomes for CBT with SM, this review demonstrates a significant lack of methodologically sound research exploring cognitive factors underpinning and maintaining SM. Case reports represented 10 studies (59%) included in this review, and data extracts were only gathered using formal methods of data collection in seven studies (41%). Data relating to cognitive factors is vital when offering evidence-based interventions based on working with cognitions and breaking maintenance cycles in order to overcome a failure to speak. As such, this review offers an initial synthesis of available evidence around cognitive factors in SM, based on participant self-report, and highlights several areas for further research.

The use of participant self-report in SM is limited, with a reliance on observer report, self-report diagnostic assessments or measures of anxiety not specific to SM (as was found in 17 excluded research articles; see Figure 2). Across the 17 studies reviewed in this paper, four studies used formal methods of self-report as the primary source of information; three of which were conducted with adults. This reflects considerable methodological challenges in gathering self-report data from participants who are unable to speak. However, the identification of five possible categories of cognitive factors in this review beyond anxiety through self-report highlights the importance of this methodology and its broad application, for example in development of theory and outcome measures specific to emotional and cognitive factors in SM. This information also has helpful implications when developing other formulations around selective mutism, beyond anxiety.

Agreements and Disagreements with Other Papers

Anxiety as a primary factor in selective mutism supports the reconceptualisation of SM as an anxiety disorder within DSM 5 (APA, 2013). Debate exists in the literature around whether SM is separate to social anxiety disorder representing a specific phobia of expressive speech (Omdal & Galloway, 2008), or falls along the same continuum (review by Muris & Hollendick, 2015). For the purpose of this review, extracted data required a specific reference to anxiety with speaking, although co-morbid social anxiety was reported in a number of studies, therefore raising issues about disentangling cognitions specific to each disorder. Fear hierarchies co-constructed with clients highlighted a number of specific fears relating to speaking aloud (Christon et al., 2012; Jackson et al., 2005; Reuther et al., 2011; Rye & Ullman, 1999). Whilst this could be considered to lend support to a hypothesis around SM as a specific phobia of speech, treatment in each study aimed to increase vocalisations, therefore situations on the hierarchy reflected treatment goals and may not indicate the absence of general social fears.

When considering a cognitive model of SAD underpinning CBT interventions, a number of parallels can be drawn between evidence-based cognitive factors maintaining SAD and emerging cognitive factors in SM. Similarities are evident between the subcategory of ‘fear of evaluation’ in SM, identified in six studies (for example predictions of others’ negative perception when the individual is unable to speak), as well as other fears such as getting something wrong; and negative beliefs about performance in a cognitive model of SAD (Clark, 2001). The concept of self-consciousness, highlighted in two studies, could also be conceptualised in the framework of processing self as a social object (Clark, 2001) within SAD. Furthermore, the category of negative self beliefs could translate to unconditional negative beliefs in SAD, although this category was only identified in five studies (29%). In addition, examples of negative beliefs and emotions (such as shame) were described as a result of mutism, which may challenge the idea of an unconditional belief, as well as raising questions about directionality of the belief (for example, questioning whether a negative self-belief preceded mutism or occurred as a consequence).

Evaluating findings in light of other theoretical approaches to SM; the category of mutism as self-protective, along with the subcategory of active control over speaking, could be understood within the framework of emotion regulation theories (Scott & Beidel,

2011). Conceptualised in this way, a failure to speak represents an active coping strategy to manage distress or difficult experiences, for example disappointment, punishment or mistrust of others (Omdal, 2007; Remschmidt et al., 2001). From a cognitive-behavioural perspective, this may represent an avoidance behaviour (or safety behaviour) which was proposed as a maintenance factor in SM by Muris and Hollendick (2015), reflecting a belief that not speaking prevents something bad from happening.

Alternatively, the cognitive factor of active control over speech could also be conceptualised as oppositionality (Hua & Major, 2016; Muris and Ollendick, 2015), through consciously choosing to withhold speech when under social pressure, or attempting to retain control. In the same way, the perception of a separate identity as mute may fit with an oppositional understanding, although all of the data extracts in this category were taken from two studies which were also represented in the active control category, therefore raising a challenge around drawing conclusions on the basis of a narrow range of data.

Behavioural inhibition theory has also been proposed as a mechanism driving a failure to speak, suggesting that during times of distress such as high anxiety; the sympathetic nervous system is activated resulting in inhibition of speech (Wong et al., 2010). Behavioural inhibition may be suitable to explain the category of lack of control over speaking, where participants reported an inability to speak despite wanting to, which may reflect an automatic biological process that they are unable to control. This theory also reflects the presence of anxiety, which is suggested implicitly (“freeze up”; Christon et al., 2012) within the category of lacking control.

The category of lacking control also reflects the idea of a reinforced pattern of behaviour that becomes hard to break. Similarly, expectations of others suggests the influence of environment in reinforcing behaviour, fitting with both behavioural explanations of learned behaviour, and developmental perspectives around the interaction between an anxious disposition and environmental factors (Wong et al., 2010).

Limitations and Biases

Despite promising connections between existing theories of SM and cognitive factors identified in this review, generalisability of findings is limited due to reliance on a small amount of data retrieved from studies of variable quality, and a lack of research

directly investigating the topic of the review. The use of self-report data in the review was an important step in increasing knowledge of cognitive maintenance factors based on direct evidence from those experiencing SM, as well as increasing reliability of evidence through avoiding a reliance on observer report. However, the reliability of self-report data around cognitions from young children particularly could be questioned due to stage of brain development where higher cognitive functions may still be developing (e.g. Blakemore & Choudhury, 2006), and the category of 'Beliefs around the reason for mutism being unknown' reflects the challenge of research with a difficulty primarily experienced in young children. The presence of several theoretical stances in relation to SM poses a challenge when synthesising data from different modalities, as the data reported is likely to be biased towards the researcher stance (for example psychodynamic). In addition, reliance on retrospective reporting of experience from adults may be subject to memory bias, with participant's understanding of why they did not speak as a child reformed over time, possibly differing from the meaning they made of mutism as a child.

The use of inductive content analysis provided a useful method of quantifying qualitative data. However, the process of identifying qualitative extracts which answered the research question and categories to cluster extracts involved a degree of subjectivity, although standardised processes were put in place to minimise risk of author bias and inter-rater reliability remained close to 80%. The degree of subjectivity and overlap in coding categories was highlighted when discussing discrepancies in coding as part of the inter-rater reliability process. This showed the role of context within each paper and the challenge of minimising interpretations and taking data extracts at face value. An alternative to an inductive approach would have been to categorise data according to a pre-determined theoretical framework of cognitions in SM. However, due to the lack of research in this area, no such framework was available.

Conclusions and Clinical Implications

In conclusion, this review provides an initial step in an otherwise unexplored area; identifying cognitive factors within SM which maintain the core symptom of a failure to speak. Categories of cognitive factors aligned with several theoretical perspectives on SM, providing additional evidence based on direct self-report. The review provided evidence of cognitive factors beyond the immediate and direct experience of anxiety. This highlights the importance of a broad assessment and formulation when working with those with SM,

in order to capture possible functions beyond anxiety maintaining a lack of speech such as self-protection and control. These cognitive factors may also highlight important considerations around implications for a young person of starting to speak, for example loss of separate identity and a sense of self, as well as increased anxiety once the coping strategy has been removed. On the basis of this review, it is recommended that research studies are conducted to directly examine cognitive factors in SM, in order to develop a cognitive model of SM which may underpin future interventions. In addition, an outcome measure for SM that moves beyond behaviour to look at emotions and cognitions (Muris & Ollendick, 2015) is important in future practice, and the cognitions identified in this review provide an initial map of possible topics to target in a questionnaire.

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Sex and Relationships Education for Individuals with Cystic Fibrosis: A Service-Directed
Approach

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Intended journal: This paper is intended for publication in the journal *Sexuality and Disability*. This journal publishes papers exploring psychological factors relating to sexuality, which fits with the scope of this paper in exploring patient experience of sex and relationships education. The journal also publishes varied types of articles beyond research, therefore would be suitable for a service-directed paper.

Introduction

Cystic fibrosis (CF) is a recessive genetic disorder caused by an abnormality of the CFTR gene, resulting in production of thick mucus particularly in the lungs and pancreas and leading to recurrent infections, breathing difficulties and digestive problems (Knott, 2015). Prevalence of CF in the UK is around 1 in 2500 births (Knott, 2015), with over 10,000 people currently living with CF in the UK (Cystic Fibrosis Trust, 2016a).

CF is life-limiting, with median predicted survival in the UK estimated at 45 years (Cystic Fibrosis Trust, 2016a), although this is increasing yearly, with a median predicted survival of more than 50 years for children born in 2000 (Dodge, Lewis, Stanton & Wilsher, 2007; Simmonds, Cullinan & Hodson, 2009). Increased life expectancy raises issues such as pregnancy and parenthood which were not previously considered possible in CF. As CF is a recessive disorder, both parents must carry the CFTR gene for a child to inherit the disorder (see Figure 1), highlighting issues around reproductive decision-making.

Background Literature

Key Sexual and Reproductive Health (SRH) Issues in CF

Beyond genetics, several other areas of SRH pose specific issues in CF. A review by Frayman and Sawyer (2015) summarised challenges around effectiveness of contraception, sexually transmitted infections (STIs), fertility, pregnancy and parenthood, and in vitro fertilisation (IVF) amongst other areas.

Around 98% of men with CF are infertile (Cystic Fibrosis Trust, 2016b). Research suggests that male infertility is often discussed at diagnosis but may not be brought up again by teams unless asked (Frayman & Sawyer, 2015). Little conclusive evidence exists for women to suggest that fertility is affected (Edenborough, 2001; Frayman & Sawyer, 2015). Despite this, Sawyer, Phelan and Bowes (1995) found misconceptions about fertility in women with CF adversely influence contraceptive use, highlighting risks of unplanned pregnancy and sexually transmitted diseases (STDs). Sawyer, Farrant, Cerritelli and Wilson (2005) found that 1 in 3 men at a clinic in Australia believed they did not need to use condoms due to being infertile, highlighting further implications for STDs, although

McEwan, Hodson and Simmonds (2012) found no difference in the prevalence of contraceptive use in adults with and without CF.

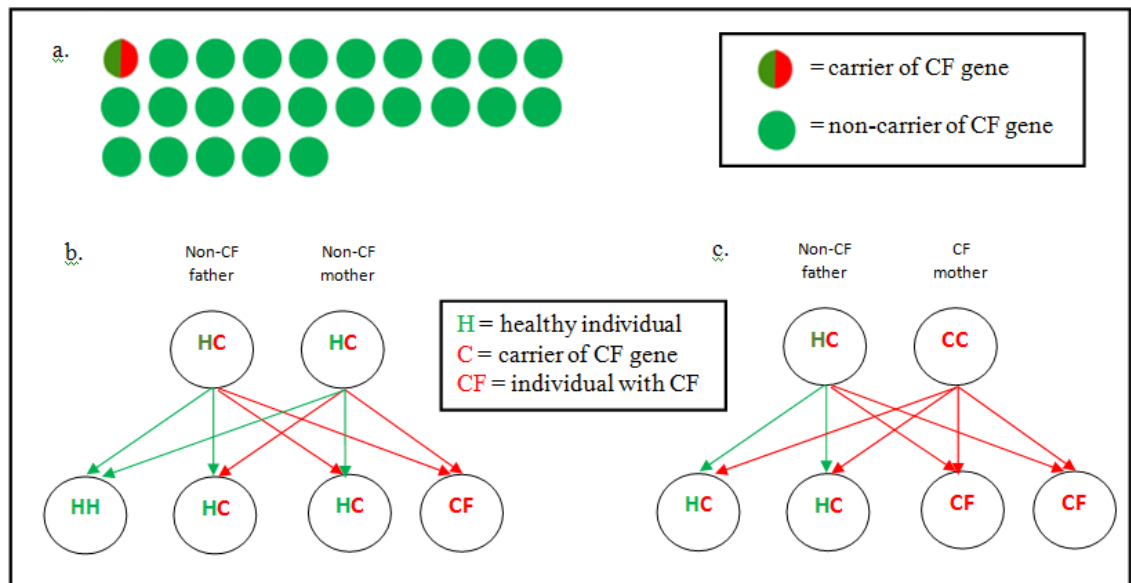


Figure 1.(a) Prevalence rate of carriers of CF gene: 1 in 25 people. (b) Genetic map for two carriers of CF gene. Probability of child having CF: 1 in 4. (c) Genetic map of one individual with CF and one carrier of CF gene. Probability of child having CF: 1 in 2. Based on information from: Cystic Fibrosis Trust (2016c).

Research has also highlighted a lack of knowledge and common misconceptions around SRH issues in CF (Frayman & Sawyer, 2015; Havermans, Abbott, Colpaert & de Boeck, 2011). Gage (2012) demonstrated a need amongst female patients for further knowledge in physiological, genetic and psychosocial SRH issues to aid reproductive decision-making. Withers (2012) also showed SRH to be a key management issue amongst adolescents with CF, suggesting it should be included in regular check-ups of adolescents' psychosocial needs. However Sargant, Smallwood and Finlay (2014) found that across several life-limiting conditions, history relating to sexual health was not being recorded for any adolescents in their sample (n=25), suggesting this does not form part of regular check-ups for adolescents, although findings may be influenced by poor record-keeping.

SRH Provision

SRH issues specific to CF raise questions about how CF services provide this information to patients. Frayman and Sawyer (2015) evaluated current SRH provision internationally, highlighting considerable discrepancies in several studies between actual

and preferred age by patients of initial discussion about SRH with CF services (with average age of initial discussions ranging from 16-17.4 years across studies; compared to preferred age ranging from 13.7-14 years). Furthermore, inconsistencies in parental and patient knowledge of SRH in CF were evident, with a high proportion wanting more information, including paediatric patients themselves (Nixon, Glazner, Martin & Sawyer, 2003).

One challenge in providing CF-specific sex and relationships education (SRE) is that evidence in some areas such as female fertility and the effects of pregnancy on women's health with CF is inconclusive and contrasting (see Frayman & Sawyer, 2015 for a review); yet factors such as these play a role in reproductive decision-making for patients with CF (Simcox, Hewison, Duff, Morton & Conway, 2009) suggesting the importance of accurate and standardised guidance. In line with this, findings of Frayman and Sawyer's review (2015) led to the proposal of a model of service provision to address unmet patient need and inconsistencies in SRH care, including a timeline of SRE for parents and patients by the CF service. Recent research published since the beginning of the present study supported findings of the review and model with qualitative data from female CF patients and programme directors in the US (Kazmerski et al., 2016). Themes included the importance of discussing SRH, with care providers initiating conversations, and barriers for patients and providers in discussing SRH, along with preferences for both written information and conversation. A limitation with the sample of programme directors was the small size (n=16) along with demographics; the majority were male with a mean age of 55 years, which may not be representative of clinicians in the CF service who would typically provide SRH information. In addition, no distinction was made between information provided in adult and paediatric services.

Context of Local Service Provision

Previous projects within the team and nationally led to the creation of a website for patients with CF around pregnancy and parenthood, however highlighted an unmet need within the paediatric service. Although paediatric clinicians routinely informed male patients about the biological effect of CF on fertility, a standardised procedure for routine SRE about issues such as contraception and sexually transmitted diseases (STDs) was not in place, despite these issues being stipulated in national legislation in schools (Department of Education, 2014). Research questions were developed to evaluate Frayman and

Sawyer's model of service provision (2015) in the UK as well as responding to the gap in current service provision in the local CF service. Before evaluating the model, accurate knowledge and understanding was needed of current service provision around sex and relationships in CF, leading to the first research question, as well as evaluating the age at which SRE might be provided within the paediatric service relative to the proposed service model, leading to the second research question. Due to service limitations and concerns around data collection directly with paediatric patients or their parents, the methodology was revised to gather retrospective information from adult patients with CF.

Aims

- To consider the age at which cystic fibrosis (CF)-specific issues within sexual and reproductive health (SRH) and relationships should be discussed and the role of the CF service in doing this
- To consider whether service provision met patients' needs according to their experience of sex and relationships education (SRE) throughout the service
- To highlight patients' needs and opinions, and enable the adult and paediatric CF services to make improvements to provision through reflecting on SRE and policies

These aims will be addressed through information gathering about SRE provision within a UK clinical population of adults with CF, in order to compare findings to those gathered in US and Australian populations (Frayman & Sawyer, 2015; Kazmerski et al., 2016) and answer further questions about the role of both adult and paediatric teams in providing SRE to patients with CF.

Research Questions

- Did past service provision in the area of SRH and relationships meet patients' needs and how this could be improved?
- What age do patients think CF-specific SRH and relationship issues should be discussed and what role does the CF service have in this?

Design

The study used a retrospective survey-based design with adult patients with CF. Initial consultation with Clinical Psychologists working across paediatric and adult Cystic Fibrosis services identified that data collection with paediatric patients or their parents would not be possible due to concerns around methodology within the paediatric service.

Method

Procedure

Phase 1: Service consultation and scoping.

Following initial scoping, phase one involved a continuing professional development (CPD) session with a Paediatric CF service around CF-specific SRH issues. Members of the multi-disciplinary team (MDT) were presented with a seminal review paper and model of SRH service provision (Frayman & Sawyer, 2015) in order to consider the application of the model and scope potential survey questions. Clinician participants took part in a feedback session facilitated by the primary researcher (see Table 1). Represented professions included psychiatry, clinical psychology, nursing and physiotherapy.

Table 1

Consultation Session Discussion Topics

Discussion topics within the consultation session
Current service provision of sex and relationships education to paediatric patients
Questions that the Paediatric CF team would like to ask patients about service provision
Concerns/queries about the research project e.g. practicalities, recruitment
Feedback about the Frayman and Sawyer model (2015) and how that might fit with current and future service provision
Initial discussion about the age at which patients might be given different information by the team

Phase 2: Questionnaire design.

Based on responses from the MDT and the literature, a 28-item survey was constructed around SRH and relationships within CF. Feedback from several adult patients

around readability and comprehension was positive. The survey received ethical approval from the University of Bath's Psychology Ethics Committee (Reference 15-243). However, revisions to the questionnaire and methodology were required to gain approval from the Research and Development (R&D) team at the hospital base; a process which took place over four months. Revisions included more multiple-choice questions and fewer free-text questions, with participants offered the option of a face-to-face interview if preferred to an online survey due to the sensitive nature of the topic. During this process, feedback on the survey was gathered through a further CPD slot with an Adult CF team. Concerns were around survey items which may not be relevant to all patients, such as urinary incontinence; and possible recall bias of specific information about age, therefore these questions were removed. Feedback from adult inpatients with CF on a respiratory hospital ward suggested that patients were comfortable with an online survey but appreciated the option of a face-to-face interview, and that the topic of SRH was important to discuss.

Once full approval was gained from the R&D department, the revised 22-item survey was piloted with four adult CF inpatients on the respiratory ward, generally receiving positive feedback. Further concerns were raised around recall, and reluctance to give critical feedback to current care providers. In response to this, retrospective questions included the response item 'don't know/can't recall' to allow for difficulties with recall, and concerns around critical feedback were addressed through the consent form, ensuring participants were aware that all data was anonymous and could be withdrawn before submission or questions could be left blank. No issues were raised with comprehension or readability.

Phase 3: Survey.

Recruitment process.

An e-mail with an advert, researcher contact details and a link to the online survey was sent to all adult patients in a UK CF service using an existing database with permission from the service. A follow-up e-mail prompt was sent three weeks later. Where e-mail contact was not possible, paper copies of the survey were sent with the advert, information sheet, consent and debriefing forms, however no copies were received back. In order to increase sample size, patients who were well enough to consent were approached to complete the survey when attending clinics at the CF service and whilst inpatients on the respiratory ward.

Participants.

Participants were adult patients (N= 223) with an open referral to a UK adult CF service. Response rate was 14%, with a final sample of 31 patients. The sample consisted of 17 males and 13 females, with a minimum age range of 18-24 and a maximum age range of 45-54. Demographic data was not received for one participant.

Measures.

A 22-item questionnaire was devised by the research team and refined through the process described above; therefore no data is available around validity and reliability. Survey questions were based on themes in the literature; refined using expert knowledge from clinicians, R&D committees and patients to determine if the survey fitted with the aims and research questions, suggesting that the survey had content and face validity. As such, qualitative data from the questionnaire was analysed largely using deductive thematic analysis in line with pre-existing themes in the literature, which reduced risk of interpretative bias by the researcher, although two inductive themes were identified.

Procedure.

Participants completed the survey on the Bristol Online Survey website by following the disseminated link. Before starting, participants were given an information sheet with researcher contact details and a statement of participation consent. Participants who consented were directed to the first question; if consent was not gained, the survey redirected to the debriefing statement. The survey included free text boxes; multiple-choice questions and grids, and was anticipated to take 10 to 20 minutes to complete. Seven questions at the end of the survey collected demographic information, including age of diagnosis. An additional question collected e-mail addresses of those keen to take part in a possible follow-up survey, however this was beyond the scope of the current project. As such, an e-mail was sent to interested participants informing them that they may be contacted at a later date by the service in order to carry out follow-up research. At the end of the survey, final consent was required as upon submission, all data was anonymised and unidentifiable, therefore participants would be unable to withdraw their data after this point. A debrief statement was given including the option to opt out of further e-mail prompts.

Results

Results from multiple-choice questions were analysed using descriptive statistics. Demographic results are summarised in Table 2.

Table 2

Demographic Summary of Sample

Category ^a	Response					
Gender	<i>Male</i>	<i>Female</i>				<i>Did not answer</i>
	54.8%	41.9%				3.2%
Age	<i>18-24</i>	<i>25-34</i>	<i>35-44</i>	<i>45-54</i>		
	41.9%	25.8%	22.6%	6.5%		3.2%
Sexuality	<i>Heterosexual</i>	<i>Gay/lesbian</i>	<i>Bisexual</i>	<i>Other</i>		
	83.9%	3.2%	3.2%	6.5%		3.2%
Marital status	<i>Single</i>	<i>Married</i>	<i>Cohabiting</i>	<i>Separated/divorced</i>		
	45.2%	29%	19.4%	3.2%		3.2%
Children	<i>Yes</i>	<i>No</i>				
	19.4%	77.4%				3.2%
Of those that said yes ^b : age of children	<i>0-4</i>	<i>5-9</i>	<i>10-14</i>	<i>15-18</i>	<i>19+</i>	
	14.2%	71.4%	0	14.2%	0	
Age of diagnosis ^c	<i>0-11m</i>	<i>1-2yrs</i>	<i>3-4yrs</i>	<i>5-6yrs</i>	<i>7-8yrs</i>	<i>20+ yrs</i>
	51.6%	16.1%	9.7%	6.5%	3.2%	6.5%

^aNo. of respondents = 30. ^bNo. of respondents = 6. ^cNo. of respondents = 29.

Paediatric Service Provision

Generally, participants agreed that advice around SRH and relationships should be provided by the CF team (Table 3), and that historically this has not been very useful (Table 4). They reported that although the largest proportion had felt able to ask the paediatric team about CF issues, the majority were unlikely to have done this (Table 5). Overall, participants would have liked more information on a range of topics from the paediatric service, suggesting an unmet need (Table 6).

Adult Service Provision

Most commonly, participants had sought information from the adult CF team around fertility and in vitro fertilisation (IVF) (Table 7). Out of 19 respondents, the majority of requests were fully met (57.9%) although a large proportion were only partially met (42.1%). Generally participants who had not sought information felt they did not need it (Table 8). Most commonly, participants rated service around SRH overall as good, followed by fair (Figure 2).

Table 3

Role of the Paediatric CF Service in SRE

Role of the Paediatric CF service ^a	Agree/Strongly Agree		Disagree/Strongly Disagree		Neither/Don't Know	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
Provide CF-specific advice	27	87.1	1	3.2	3	9.7
Provide general advice	21	67.7	4	12.9	6	19.4
Provide updates	16	51.6	5	16.1	10	32.3
Not provide SRE without being asked	7	22.6	14	45.2	10	32.3
Play a role in discussing SRH with partners	19	61.3	4	12.9	8	25.8

^aNo. of respondents = 31

Table 4

Support around SRH

Source of support ^a	Very useful		Fairly useful		Not very useful		Don't know/can't remember	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
School	3	9.7	12	38.7	14	45.2	2	6.5
Parents	3	9.7	16	51.6	10	32.3	2	6.5
Paediatric CF service	2	6.5	9	29	14	45.2	6	19.4

^aNo. of respondents = 31

Table 5

Information Requests to the Paediatric CF Service

Question	Response	<i>n</i>	%
Able to ask the team about SRH ^a	Yes	12	40
	No	9	30
	Can't recall	9	30
Likelihood of asking the team about SRH ^b	Very/fairly likely	8	27.6
	Very/fairly unlikely	19	65.5
	Don't know/can't remember	2	6.9

^aNo. of respondents = 30. ^bNo. of respondents = 29.

Table 6

Information Provision by the Paediatric CF Service

Topic of information ^a	Got all the information needed		Would have liked more information/support		Didn't want this information/support		Not sure/can't remember	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
Contraception	6	20	12	40	7	23.3	5	16.7
IVF	2	6.7	15	50	7	23.3	6	20
Pregnancy	5	16.7	13	43.3	7	23.3	5	16.7
Fertility	4	13.3	16	53.3	5	16.7	5	16.7
Parenthood	3	10	15	50	5	16.7	7	23.3

^aNo. of respondents = 30.

Table 7

Topics of Information Sought from the Adult CF Service

Topic ^a	<i>n</i>	%
Contraception	6	33.3
IVF	10	55.6
Fertility	12	66.7
Pregnancy	8	44.4
Parenthood	5	27.8

^aNo. of respondents = 18.

Table 8

Rationale for Not Seeking Information

Reasons for not seeking information ^a	<i>n</i>	%	Comments
Haven't needed the information	10	71.4	-
Don't feel I can ask the team	1	7.1	-
Prefer other sources	4	28.6	-
Other:	2	14.3	"Because I took the decision into my own hands and sorted it out before they spoke to me about it all because I knew already how can affect you with cf" / "Internet"

^aNo. of respondents = 14.

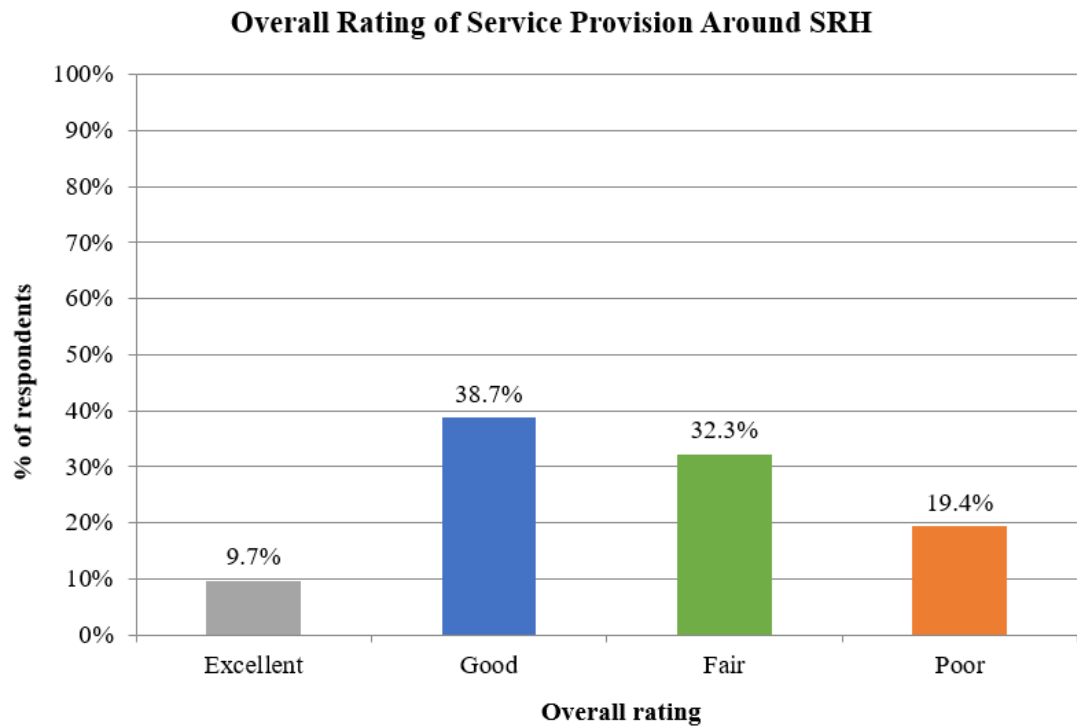


Figure 2. Participants' overall rating of provision by the CF service around SRH. No. of respondents = 31.

Thematic Analysis of Qualitative Responses

Results from free-text questions were analysed predominantly through deductive thematic analysis, as themes were anticipated based on previous research findings that shaped questions, although some inductive analysis occurred to capture themes not predicted by the literature. Fourteen initial themes were refined to eight (see Table 9) through a process of re-reading responses and codes, and returning to the research question and literature. Within each theme, responses referring specifically to adult or paediatric services were identified.

Table 9

Final Themes from Qualitative Data

Final themes	
1	Insufficient information and awareness about SRH in CF
2	Barriers to discussing SRH
3	Individual differences
4	Roles and responsibilities of the CF team
5	Content
6	Age and time
7	SRH as part of CF overall
8	The use of other sources of information

Theme 1.

Insufficient information and awareness about SRH in CF.

Several participants reported that they “*did not receive any information in paediatrics*” and suggested that receiving more information “*would have been useful*” (participant 16).

“*I didn’t even know there might be issues until I heard about it in a year 10 science lesson*” (participant 19)

Some responses suggested frustration at a lack of information:

“*I would like to point out that the ‘hospital’ response above would be more accurate with a box for: non-existent.*” (participant 16)

Other responses highlighted the negative emotional impact of not receiving sufficient information:

“At age 21 I was tested for fertility... No one really ever spoke to me about the results (infertile). I suspected I should just accept it and get on with it... I spent too long thinking the worst.” (participant 19)

“I would have loved to receive any information about sexual issues in my early teens; it simply wasn't discussed. Much to my detriment.” (participant 7)

“...you don't realise how much it affects you until you start and go through it all, worrying about things you shouldn't and would be solved if the cf team spoke more about it all” (participant 17)

One participant when reflecting on the outcome of tests with the clinic wrote: *“It was almost as if the result was an after thought... on reflection this was a huge moment in my life and could have been dealt with better” (participant 19)*

Theme 2.

Barriers to discussing SRH.

Participants highlighted key barriers such as being *“Too embarrassed (sic) to raise any issues” (participant 7)* and feeling it was *“awkward to bring up” (participant 1)* that prevented them from having conversations about SRH with the CF team.

In particular, they highlighted a desire for the CF team to initiate discussions to overcome barriers:

“It should be raised by the staff...many folk are likely to be embarrassed to raise it themselves” (participant 16)

One participant highlighted the barrier of cultural background:

“...coming from a middle class background it wasn't the done thing.” (participant 7)

Another highlighted a potential barrier of trust in the CF team:

“...personally I have to trust that person I couldn't go to a consultant but a cf nurse are always the best” (participant 17)

Theme 3.

Individual differences (inductive).

Another interesting pattern in the data was the influence of individual differences, highlighting that *“every individual patient is different”* (participant 15) and that *“... Professionals should gauge the individuals needs and respond appropriately.”* (participant 19). This included optionality, with patients suggesting *“it would be good to OFFER CF related sex/relationships advice so it is there if someone needs it”* (participant 3) but *“if the patient does not want to discuss it, they shouldn't have to”* (participant 5). This also included considering different formats for information, for example taking *“... a leaflet in a clinic room which gave advice.”* (participant 18), as *“the option of written information may be preferred by some.”* (participant 5).

Theme 4.

Roles and responsibilities of the CF team.

Several participants highlighted the CF team's role in developing a supportive relationship with patients as well as providing information.

“it's important to have the cf team by your side ever (sic) step and so you both have a understanding of what you want in your life” (participant 17)

“I feel i have a large knowledge base & should i have any queries i can count on my CF team to be there for me.” (participant 8)

Other participants highlighted the responsibility of the CF team to offer *“an open and honest platform for discussion around these topics... Doors for conversation should always be left open.”* (participant 19), with patients knowing that information *“is important and freely available, judgement-free”* (participant 15).

Several participants suggested a role in moving beyond medical information and perspectives, offering *“Good advice from a personal point of view not a medical one.”* (participant 20) and recognising that *“Psychological health is a huge part of cystic fibrosis”* (participant 19), for example psychologists helping *“to get you in a place mentally in preparation for IVF and becoming a parent.”* (participant 8).

Theme 5.

Content.

Participants highlighted when the adult CF team had helpfully provided information on topics such as “*information about IVF, pregnancy and the risks to lungs during my late 20's.*” (participant 18). Other topics where further information was required included: “*a little bit more info on the IVF front.*” (participant 25); “*...a federal to be tested for fertility*” (participant 1); advice about “*going on contraception and explain how pregnancy can affect you*” (participant 17); “*testing partners for cf gene*” (participant 12) and “*General help with conception if you want to get pregnant.*” (participant 20). One participant suggested that “*... the subject of sex, different positions and coping with CF was never really brought up.*” (participant 18).

Some participants suggested that they would have liked “*A general intro to cf related issues...Maybe at annual review a generic question offering support in this area*” (participant 14). Others highlighted content that they had been unaware of or where information was assumed to be known:

“*There are assumptions made as an adult you are aware about sex and suitable positions to assist with lung conditions. However, this is not true*” (participant 18)

Theme 6.

Age and time.

A number of participants highlighted the need for information provided to be “*age appropriate.*” (participant 7), suggesting that “*...the right care at the right age can make all the difference*” (participant 18).

Some participants suggested that information did not apply to them whilst in paediatric services:

“*I did not come close to being sexually active until I was 19*” (participant 15)

“*issue of sexual health was not an issue at just 16...more important at adult clinic than paediatric.*” (participant 12)

Others highlighted the challenge of providing standardised information at the same age to all patients, linking back to the theme of individual differences:

“...Some become sexually active earlier than others, which makes it difficult to generalise the timing of the delivery of such information.” (participant 15)

“No one should be left without the answers they need but at the same time no one should be overloaded with information they might struggle to process at a young age” (participant 19)

Participants also highlighted changes over time in the discussion of SRH in the CF service, with one participant stating: *“I felt like it was not appropriate or indeed back then the hospitals role” (participant 14)*, whereas others saw the lack of information in the past as a missed opportunity:

“I would have loved to receive any information about sexual issues in my early teens; it simply wasn't discussed” (participant 7)

“... 25 + years ago this was not even considered... just did not exist whilst growing up through peads [sic] service” (participant 14)

Theme 7.

SRH as part of CF overall (inductive).

The position of SRH as an important part of the broader picture of CF was not anticipated from previous research. Participants suggested that *“it's important to know about everything because of your illness” (participant 17)*, including SRH in order to *“have more of a full understanding on my disease” (participant 16)*.

One participant considered SRH simply as one part of their overall CF, suggesting that *“guidance from the hospital are purely how CF deviates me from the norm, and this stretches to most aspects of my adult life.” (participant 15)*, whereas others placed SRH in a position of high importance within their overall care: *“Psychological health is a huge part of cystic fibrosis” (participant 18)*.

In contrast to the rest of the theme, one participant made a clear distinction between SRH and other aspects of CF:

“clinic was to talk about lungs and digestion, not sex” (participant 16)

Theme 8.

The use of other sources of information.

A number of participants highlighted that rather than the CF team being responsible for SRH, sources such as *“parents should be responsible for educating their children on sex and reproduction” (participant 2)*, as *“General sex/relationships advice is supplied by GPs, school sessions, school nurses, adverts, online etc so I don't think it's necessary on top of this to have it in CF Clinic!” (participant 3)*.

Other sources of information included school:

“I heard about it in a year 10 science lesson” (participant 19)

Media, for example TV programmes such as *“Embarrassing bodies” (participant 17)* or *“online info” (participant 29)* from *“...the internet...when it became more prevalent at home.” (participant 16)*

Other medical professionals, such as *“Doctors” (participant 21)* and *“Gynaecology” (participant 17)*.

One participant highlighted the informal distribution of information around SRH via peers:

“It was mostly friends, rumour and suggestion.” (participant 16)

Discussion

Results generally supported previous findings around insufficient and variable service provision outlined in Frayman and Sawyer’s review (2015). Patients reported a lack of information provision in paediatric and adult services, which in some cases had resulted in negative effects such as spending *“too long thinking the worst”* about being unable to have children due to infertility (participant 19). This highlights psychological consequences of insufficient service provision around SRH, in addition to other risks such as reduced contraceptive use (Etherington, Huntington, Conway & Peckham, 2012; Sawyer et al., 2005), thus supporting the importance of a standardised approach.

In response to the first research question, findings suggest that paediatric provision generally did not meet service user need. Participants most frequently described it as ‘not very useful’, reporting that they wanted more information on a range of SRH topics. This supports evidence directly from paediatric patients (Nixon et al., 2003), as well as parent-reported knowledge from paediatric services (Frayman, Cerritelli, Wilson, & Sawyer, 2008) suggesting a reliable finding despite a retrospective approach to data collection. Barriers to information seeking such as embarrassment and a desire for the CF team to initiate conversations highlight the role of the CF service in meeting patient need in this area. Patients did not feel that information provision should be dependent on a patient request, but should be provided as standard within the service, including refreshers of information over time, in line with Frayman and Sawyer’s model (2015) of service provision.

In contrast to previous research, some respondents did not feel that SRE was relevant for them whilst in the paediatric service, with reasons including the age at which they became sexually active. This highlights the importance of sensitivity to individual differences when providing SRE. In line with this, previous research has advocated an individualised approach in terms of age and format (Kazmerski, 2016; Withers et al., 2012), however suggests that giving patients the option of SRH information is still necessary.

Findings indicating that patient requests for SRH information were fulfilled less than two thirds of the time highlight a shortfalling in service provision even when patients overcame identified barriers to initiate the conversation. Some participants reported not seeking information from the adult service as they did not require the information on certain topics. Based on the demographic spread, where participants most commonly fell in the 18-24 age range and did not have children, it could be hypothesised that the proportion of patients seeking information in the future may increase as topics become applicable to them, for example the two most commonly requested topics of fertility and IVF. In addition, findings in theme 7 that patients tended to view SRH as an important part of their overall understanding of CF emphasise the importance of prioritising service provision in this area. Furthermore, one fifth of participants reported seeking information from other sources rather than the CF team. Barriers identified by participants to seeking information may lead them to use other sources, therefore reducing these barriers, as well as increasing

availability of information, may increase the level of information sought and the prevalence of discourse around SRH in services.

Service Implications

Implications for health service practice suggest a need for improvement in consistency and overall provision of information about SRH, particularly in a paediatric setting. Patients also reported that refreshers of information would be beneficial, suggesting that information provision should be ongoing to fit with patient need, rather than a one-off provision. In addition, findings highlighted an important role for the CF service, particularly the paediatric team, in developing a strong relationship with patients and instigating conversations about SRH issues in order to overcome barriers facing patients in discussing these issues, such as embarrassment and awkwardness.

Findings and recommendations were presented to the paediatric CF team during an MDT meeting and disseminated to the adult CF service where comments were invited. Feedback was mixed, with interest expressed in how to implement findings into the service, and ongoing concerns raised about the role of the paediatric service in communicating information to patients, and possible memory bias in the study. Some clinicians felt delivery of sex education was already in place; however staff acknowledged that this was not consistent or standardised, and was not prioritised within the service. There was some discussion about whether to provide information to parents of paediatric patients or adolescents themselves, with the general consensus being that it may be helpful to consult both, and it was agreed that a consultation would be set up with parents as a consequence of study findings. This is in line with a hypothesis that parents may want more information around SRH from the CF service in order to support their children, as was found in Frayman et al., 2008. It was proposed that the format may be a parent information evening held around the topic of SRH, as these are already held in the service. The paediatric team aim to agree a protocol as to how to deliver information around SRH to adolescents. In addition, they were keen to identify further training or information about relevant issues in SRH for their patients, due to not feeling fully informed about all issues. The team began to research potential resources such as leaflets in response to the identified information need for paediatric patients. Clinical psychologists within the team have agreed to be responsible for organising the next step of consultation with parents, as well as considering resources for increasing knowledge/training. The process of conducting the

project and feedback from the team highlighted the challenges for services in talking about sex, including a lack of confidence in knowledge and competence in initiating these discussions and the taboo that this topic still carries. This suggests the importance of additional training in services and continuing discourse to address these challenges.

Limitations

Reliance in this study on self-reported retrospective information from adult patients may reduce reliability due to difficulties with recall and memory bias. Sample size was small (n=31), limiting generalisability, and the sensitivity of the research topic may have created a bias towards participants who felt more able to discuss SRH or those with strong views on the topic, as well as a possible bias in answers given in line with concerns about criticising a current care provider. As such, results may not be representative of all patients with CF.

Conclusions and Further Research

This study contributes qualitative information about service provision within paediatric and adult CF services in the UK. Despite reliance on retrospective data, the theme of 'age and time' indicated that retrospective reflection enabled some participants to consider information that would have been useful with hindsight, that they may not have realised they needed at the time. This is therefore a useful finding as a result of retrospective methodology. Further research could explore the information needs of both paediatric patients and their parents, enabling data to be collected on the specific age of provision. In addition, research around overcoming barriers to information seeking by patients and provision by clinicians would build on the work conducted by Kazmerski et al. (2016). Finally, further research could explore the format and frequency of SRE provision, building on findings around refreshers and the usefulness of written information. These questions could be answered through consultation with patients, perhaps in the form of interviews or electronic focus groups, for example through video calls, to overcome the risk of cross-infection when patients meet in person. The possibility of further research as recommended has already been facilitated through the collection of e-mail addresses within the current study for patients interested in being involved in future interviews in this area.

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The Relationship between Challenging Behaviour and Social Story™ Interventions: A
Pilot Study in a Naturalistic Setting

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Intended journal: This paper is intended for publication in the Research in Developmental Disabilities journal. This journal publishes empirical research studies which contribute to an understanding of developmental difficulties, including intervention studies. The current paper contributes to an understanding of an effective intervention for young people with Autism Spectrum Disorder, making the journal highly appropriate for dissemination of the article.

Literature Review

First described by Kanner in 1943, Autism Spectrum Disorder (ASD) is defined in *The Diagnostic and Statistical Manual of Mental Disorders* (5th ed.; American Psychiatric Association, 2013) as “persistent deficits in social communication and social interaction across multiple contexts” (p. 50) and restricted and repetitive behaviour, present since childhood and causing significant impairment. Characteristics vary across the spectrum, typically including repetitive speech, movement and play; preference for routine and difficulty understanding social situations (NHS, n.d.). Prevalence of ASD diagnoses is typically higher in males, with a male:female ratio of 4-5:1 (Fombonne, 2009). Additionally, up to 50% of young people with ASD have an associated intellectual disability (NHS, n.d.).

Theories of Challenging Behaviour

ASD can lead to frustration and anxiety as children struggle to understand situations and express their feelings (Autism Speaks Incorporation, 2012; U.S. Department of Health and Human Services, 2012). This often manifests as ‘challenging behaviour’, defined as “behaviour that challenges – whether it is a challenge to our understanding, our own well-being or our child’s or else to our ability to carry out our responsibilities as parents or professionals” (Whitaker, 2001; p.4). Behaviours can include disruption, aggression, self-injury, and repetitive behaviours (Horner, Carr, Strain, Todd & Reed, 2002; National Autistic Society, 2015), and can place the safety of the child and others at risk (Emerson & Einfeld, 2011), thus identifying it as a necessary target for intervention.

Researchers have theorised several biological, behavioural and social determinants for challenging behaviour (Emerson & Einfeld, 2011). Behavioural theory proposes that behaviour serves a function for the individual, shaped by environmental consequences (operant conditioning; Emerson & Einfeld, 2011; Skinner, 1938) and maintained by positive reinforcement (obtaining a desired consequence/object) or negative reinforcement (removing or preventing an undesirable consequence/object; Emerson & Einfeld, 2011; Skinner, 1938). It occurs in a functional relationship; triggered by antecedents with consequences that determine reinforcement or extinction (Skinner’s Stimulus-Response-Stimulus model, 1938; Dixon, Vogel, & Tarbox, 2012). Research highlights four categories of function: demand avoidance, accessing attention, accessing

tangibles/activities and sensory stimulation (Iwata, Dorsey, Slifer, Bauman, & Richman, 1994; Tarbox et al., 2009). Function is identified through functional assessment (FA; Iwata et al., 1994; Harrower, Fox, Dunlap, & Kincaid, 2000), which informs intervention methods, such as pivotal response treatment (Koegel, Koegel, Harrower & Carter, 1999). Other examples include functional communication training (FCT) based on FA, shown to successfully decrease challenging behaviour in line with the communication hypothesis (that behaviour functions as nonverbal requests for social consequences; Carr & Durand, 1985).

Social Stories™

Not all effective interventions for challenging behaviour have been developed based on operant principles. Social Stories™ (Gray & Garand, 1993) are stories about a social situation, skill or behaviour, written by professionals or parents and including the child's perspective. The stories aim to improve social skills and understanding by describing social concepts and situations using "relevant social cues, perspectives, and common responses" (Gray, 2004, p. 2).

Evidence for Social Stories

Social stories have also been shown to reduce challenging behaviour (e.g. Kokina & Kern, 2010) through describing an adaptive way to fulfil its purpose. Gray suggests that reduced challenging behaviour occurs indirectly through changes in cognitive deficits such as theory of mind and central coherence (2004; see Table 1 for details). However, Reynhout and Carter (2011a) highlighted a lack of evidence, suggesting that social stories function as contingency contracts through outlining preceding events that necessitate a behaviour, expected behaviour from the individual, and likely consequences, or "natural reinforcers" (p.375).

Evidence for Factors Influencing Effectiveness

According to the creator of Social Stories, valid social stories must conform to published guidelines, including a specific ratio of sentence types (Gray, 2004). These have been updated several times (see Table 2 for a comparison and examples of sentence types in Appendix C1; Gray, 1998, 2004; 2015; Sansosti, Powell-Smith, & Kincaid, 2004); most recently conceptualised as descriptive and coaching sentences (Gray, 2015). However,

research has questioned the empirical basis for this ratio (Kuoeh & Mirenda, 2003; Reynhout & Carter, 2006), showing that stories with more directive (coaching) sentences (Reynhout & Carter, 2006) or only directive sentences (Quirnbach, Lincoln, Feinberg-Gizzo, Ingersoll, & Andrews, 2009) can be equally or more effective than those conforming to guidelines (Kokina & Kern, 2010). A survey-based study of teachers in Australia (Reynhout & Carter, 2009) found that 57% always or sometimes adhered to Gray's guidelines, and 36% were unsure; 66% of sample social stories either did not fulfil social story criteria or were uncodable.

Table 1

Theoretical Underpinnings to Social Stories Proposed by Gray (2004)

	Definition	References	Relevance to social stories	References
Theory of mind	Attributing mental states to self or others	Baron-Cohen, Leslie & Frith, 1985; Premack & Woodruff, 1978	Considerable research has demonstrated a deficit in this ability to take perspectives of others in autism, and these perspectives are clearly outlined within social stories.	Baron-Cohen, 2001; Baron-Cohen et al., 1985
	Drawing together individual aspects within a context to understand the bigger picture.	Baron-Cohen, 2001; Frith & Happé, 1994	Social stories outline important social information carried in these forms of unspoken communication in a clear and predictable way, thus reducing challenging behaviour that can result from deficits in social understanding.	Gray, 2004

Table 2

Social Story Guidelines

Social Story Guidelines (Gray, 2004)	Social Story Updated Guidelines (Gray, 2015)
<ul style="list-style-type: none"> • Social stories should always include descriptive sentences and may also include five other sentence types (see below). • Directive and control sentences should be limited in each story. It may not be necessary to include directive sentences in every story. • Sentences should fit into the “Social Story Formula” (p.21), equivalent to a ratio of one directive/control sentence for every 2-5 other sentences (Gray, 1998, 2004; Sansosti et al, 2004). 	<ul style="list-style-type: none"> • Social stories should always include descriptive sentences and may also include coaching sentences. • Social stories should describe more than they direct. • Sentences should fit into the Social Story Formula, which can include an unlimited number of descriptive sentences but no more than one coaching sentence for every two or more descriptive sentences.

Treatment Response to Social Stories

Multiple reviews have evaluated treatment response to social stories. A descriptive review by Sansosti et al. (2004) found social stories to be effective in decreasing challenging behaviour and increasing social skills for individuals with ASD; however critics questioned the validity of a descriptive approach and reliance on single case studies. Karkhaneh et al.’s review (2010) also demonstrated effectiveness of social stories in 5 out of 6 comparative trials, although methodological quality was questioned as all studies were unpublished dissertations.

Several meta-analyses have used statistical methods to quantify effectiveness (reviewed by Kokina & Kaczmarek, 2013; see Table 3). However, a review by Leaf et al. (2015) found that 92.7% of single case designs could not draw causal conclusions due to methodological limitations, highlighting challenges in assessing effectiveness within the literature. Research is limited into moderators of treatment response (Hutchins & Prelock, 2014). Proposed moderators include intervention target, with higher effectiveness for reducing challenging behaviour than increasing social behaviour (Kokina & Kern, 2010) and the use of comprehension checks (Reynhout & Carter, 2006), although research has shown effectiveness for nonverbal children and those with intellectual disabilities

(Hutchins & Prelock, 2014; Kim, Blair & Lim, 2014). Kokina and Kern (2010) showed an association between FA and treatment response (average percentage of non-overlapping data; PND of 86% compared to 53% for those not using FA); however, reliability is questionable due to the small number of studies. A recent study by Pane, Sidener, Vladescu, and Nirgudkar (2015) used FA in an alternating treatment design, comparing effectiveness of function-based social stories; non-function-based social stories and social stories describing baseline. Results supported effectiveness of function-based stories alongside FCT in reducing challenging behaviour and increasing alternative behaviour compared to other study conditions. However, the differential effect of the social story was unknown due to the addition of FCT, and sample size was very small (n=2).

Table 3

Outcome of Meta-Analyses Reviewing Treatment Response to Social Stories

Statistical method	Percentage of non-overlapping data (PND; Scruggs & Mastropieri, 2013)				Percentage exceeding the mean (PEM)
Effectiveness	91-100 = highly effective	71-90 = moderately effective	51-70 = mildly effective	0-50 = non-effective	Moderate effectiveness
References	No papers	No papers	Kokina & Kern, 2010; Reynhout & Carter, 2011b; Sani Bozkhurt & Vuran, 2014	Reynhout & Carter, 2006; Test, Richter, Knight & Spooner, 2011	Reynhout & Carter, 2011b

Functional Assessment in Social Stories

Preliminary findings into FA (Kokina & Kern, 2010; Pane et al., 2015) are in line with evidence for other challenging behaviour interventions (Emerson & Einfeld, 2011; Horner et al., 2002; Martinez, Werch and Conroy, 2016) and recommendations to gather information prior to social stories (Gray, 1998). This could have implications for improving consistency of treatment response, by tailoring social stories to address specific triggers and reinforcers according to behavioural function (in line with Pane et al., 2015).

Without FA, social stories may inadvertently reinforce behaviour by fulfilling the desired function; or be ineffective by addressing the incorrect function (Pane et al., 2015). It is therefore hypothesised that treatment response would vary according to function of the targeted behaviour and how closely the intervention matches that function (labelled functional congruence, FC; O'Neill et al., 1997).

This study therefore aimed to explore function of challenging behaviour and functional congruence as possible influences on treatment response to a social story intervention in a naturalistic setting. A naturalistic setting allowed exploration of applicability of social story guidelines and research findings in the real world (Reynhout & Carter, 2006; Rust & Smith, 2006), fitting with recent recommendations by Martinez et al. (2016) for teachers to deliver interventions within research to assess feasibility and practicality. The setting also contrasted to Pane et al.'s study (2015) where conditions were manipulated experimentally, reducing ecological validity. Marshall et al. (2016) recently conducted a feasibility study for a randomised control trial into effectiveness of social stories within naturalistic settings. The study employed questionnaires and diary measures to gather data around challenging behaviour (Marshall et al., 2016), supporting methodology in this study. However, several variables were not explored such as comprehension or change in social skills; both of which will be tentatively explored in the present study, along with conformity to Gray's sentence ratio (2015).

Research Questions

- Does the function of challenging behaviour in children with ASD influence treatment response to a social story intervention?
- Do social story interventions influence social skills over a short period of time?
- Does social story structure influence treatment response to a social story intervention?

Hypotheses

1. Treatment response of a social story intervention targeting challenging behaviour among children with ASD will be influenced by behavioural function.
2. There will be a higher proportion of positive treatment response among participants where the social story is functionally congruent with behaviour than for those where function targeted in the social story does not match behavioural function.

3. For participants where the social story and behaviour are non-congruent, there will be a negative treatment response (see operationalisation below).
4. Functional congruence of a social story and behaviour will influence treatment response above concordance of the social story to a standardised sentence ratio
5. Participants will show an improvement in social skills following a social story intervention, as assessed by pre and post measures of social skills.

Method

The study occurred in collaboration with a wider social stories project run by Professor Mark Brosnan, field supervisor and Dr Liz Smith; acting as a pilot into the variable of 'behavioural function'.

Ethical Approval

Ethical approval was gained from the Psychology Department Ethics Committee at the University of Bath prior to the study commencing (Reference 16-071). Informed consent was gained from parents of participants and educational staff who completed questionnaires via information sheets and consent forms, and assent was gained from participants where possible.

Sample Size and Power

Power was calculated based on analysis into functional congruence and treatment response. Previous studies into functional congruence and challenging behaviour had sample sizes of four (Cihak, Kildare, Smith, McMahon, & Quinn-Brown, 2012) and two (Pane et al., 2015). As such, power was calculated a priori using estimates of effect size based on similar research findings (see Table 4).

Power analysis using G*Power suggested a sample size of 54, with 27 in each group (two-tailed; power =0.8; α =0.05; effect size for functionally congruent =0.8; effect size for non-congruent =0.4; allocation ratio =1). However this target was based on experimental studies, therefore was challenging to reach in the current pilot study using a naturalistic setting, with a difficult population group for recruitment. This was reflected in the final sample size of 16; the study was underpowered and parametric tests were not possible.

Participants and Recruitment

Participants were 16 children (13 male; 3 female) with a primary diagnosis of ASD, aged between 5 and 13 years ($M=9$ years; $SD= 2.31$; see Table 5 for demographic information). To maintain anonymity, demographic information was separated from specific behaviours and comprehension levels. Behaviours were summarised into five broad categories (see Table 6). A purposive sampling approach was used within schools in the South West with a specialism in ASD or special educational needs, identified through map searches and researcher knowledge (see Figure 1). Approximately one third of school visits ($n=4$; 36%) during recruitment took place with schools who did not subsequently participate. The final sample was recruited from five schools across 10 classes (see Figure 2). Table 7 outlines inclusion/exclusion criteria for participants.

Table 4

Research Basis for Effect Size

Research Basis for ES*	Intervention	No. of studies reviewed	Categorised by FC**?	Overall PND*** (ES)	Adjustments
Reynhout & Carter (2011b)	Social Stories™ for challenging behaviour	26	No	48	No account of functional congruence – round down to 0.4
Kokina & Kern (2010)	Social Stories™ using a priori functional assessment	3	No	86	Small no. of studies – rounded down to 0.8

*ES=Effect Size **FC=Functional Congruence ***PND=Percentage of Non-Overlapping Data

Table 5

Demographic Characteristics of Sample

Participant no.	Gender	Age (years)
001	M	6
002	M	5
003	M	7
004	M	6
005	F	10
006	M	10
007	M	9
008	F	9
009	M	9
010	M	13
011	M	12
012	M	6
013	F	7
014	M	11
015	M	9
016	M	9

Table 6

Categories of Target Behaviours

Category of target behaviour	No. of participants
Inappropriate vocalisations	2
Physical contact/aggression towards others	5
Self-directed aggression (including destroying own property, self-injurious behaviour)	4
Inappropriate sexual behaviour	2
Impulsive behaviour to fulfil needs	3

Table 7

Screening Criteria for Study Participants

Inclusion criteria	Exclusion criteria	Conditional criteria
Participants with a primary diagnosis of ASD	Individuals with any other primary diagnosis than ASD.	Comprehension was included as a conditional criterion based on educational staff deeming that social stories were suitable for the child given their comprehension level.
Displaying an observable challenging behaviour in school (target behaviour).	Unable to identify a suitable target behaviour according to inclusion criteria.	Rationale: Schools currently use social stories with children of varying abilities, however, research has indicated the possible influence of comprehension on effectiveness of social stories (Kokina and Kern, 2010).
Target behaviour was rated by educational staff as fulfilling a minimum frequency of at least 50% of the time in school according to the ASD-Behaviour Problems for Children (ASD-BPC). A score of 3 on the ASD-BPC equalled half the time.		Comprehension was assessed according to academic records of national curriculum levels), ranging from P3i to 2B (median = P6-7; mode = P6), with 10 participants (62.5%) assessed at below national curriculum levels (P levels). Comprehension was included in analysis after data collection as a possible confounding variable.

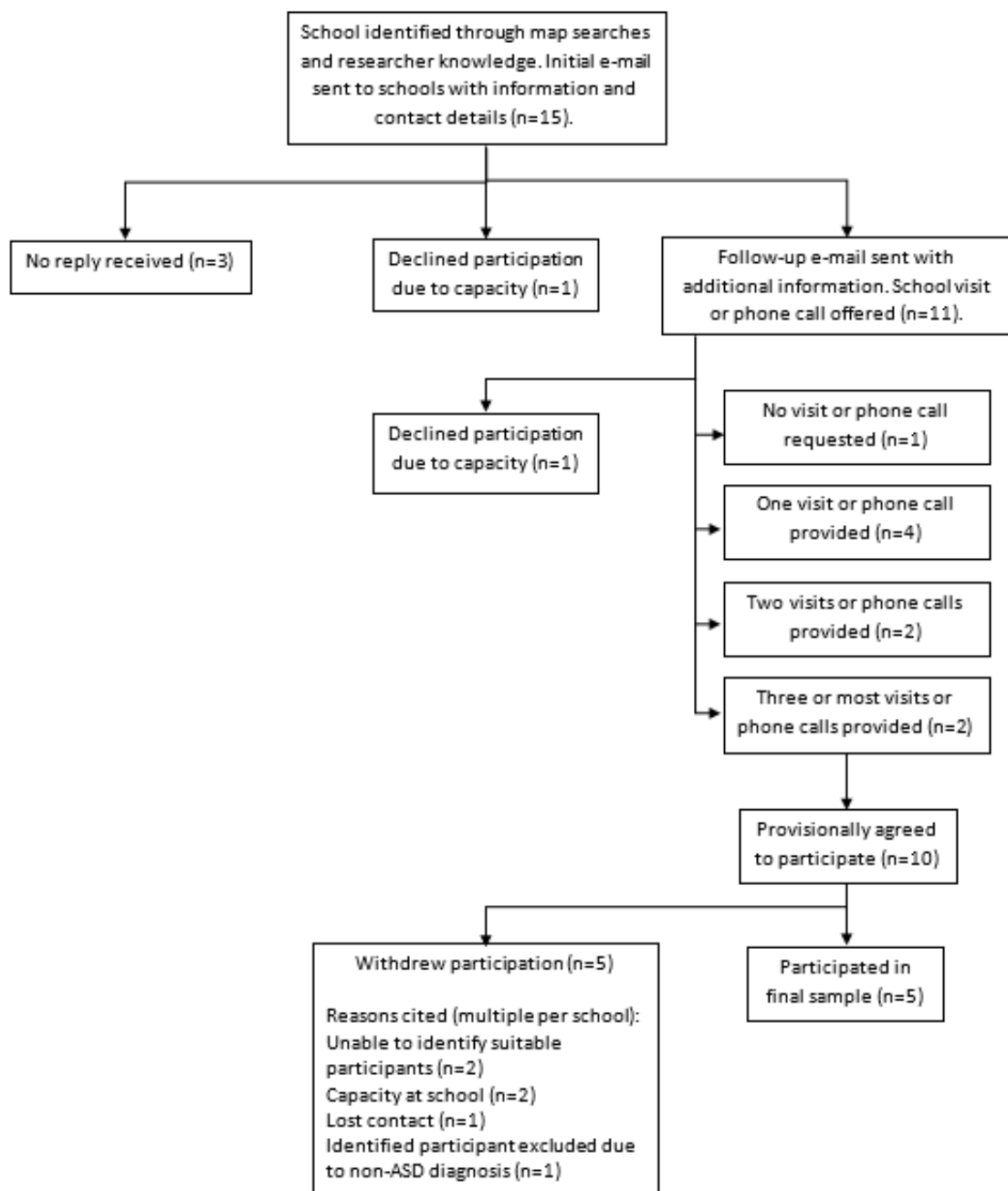


Figure 1. Recruitment process for schools.

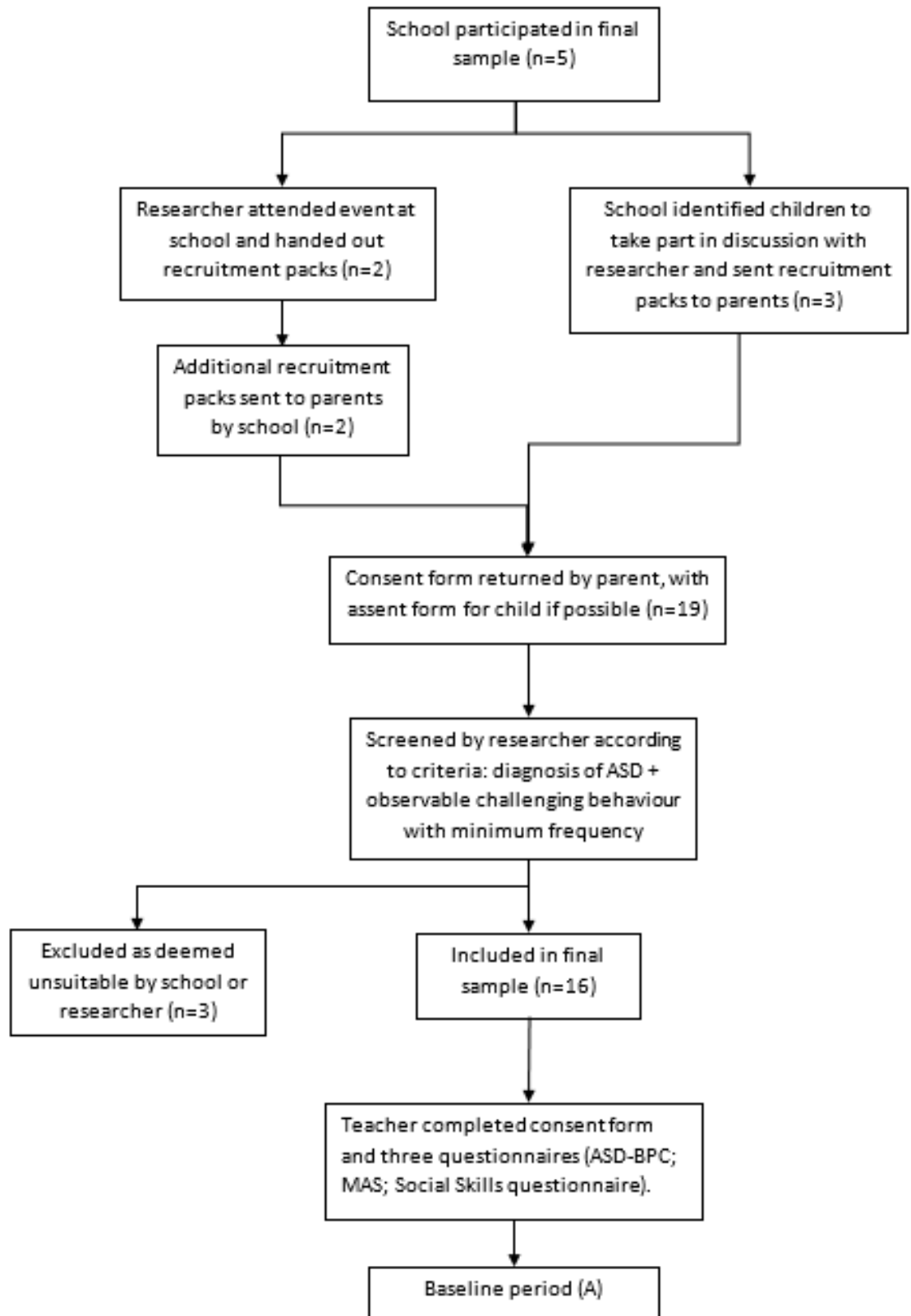


Figure 2. Recruitment process for participants within each school setting.

Measures: see Table 8.

Service user participation shaped measures created for the study. A class of children with ASD gave input on the young person's feedback form, leading to changes in formatting of the first and last questions. Additionally, feedback from teachers at the first participatory school led to simplified formatting of behaviour recording forms, to increase feasibility of completion within classrooms.

Table 8

Measures used in the Pilot Study

Target of measure	Name of measure	Author and date	Reliability and Validity
Challenging behaviour	ASD-Behaviour Problems for Children (ASD-BPC)	Matson, Gonzalez and Rivet, 2008	Inter-rater reliability = .49; test-retest reliability = .64, internal consistency = .90.
	Motivation Assessment Scale	Durand & Crimmins, 1988	Good inter-rater and test-retest reliability (correlations significant at .001 level) and high predictive validity
Social skills	Social Skills Questionnaire-Teacher	Spence, 1995a	Internal consistency = .96; construct validity supported through association between scores on SSQ-T and peer ratings (Spence, 1995b)
Baseline period observations	Behaviour record form	Created by the research team for the study (see Appendix C2)	N/A
	ABC charts		N/A
Participant feedback	Young Person Feedback Form	Created by the research team for the study (see Appendix C2)	N/A

Procedure

Figure 3 provides a detailed outline of study procedure. Each period took place within the classroom for 5 school days (or ten sessions, with one session defined as the period before lunch or after lunch).

Phase 1: Functional assessment.

A target behaviour was identified through discussions with educational staff, supported by the ASD-BPC. Function on the MAS was validated for 50% of participants via direct functional assessment by researchers using observational ABC charts.

Consistency between the two measures was 75% for at least one function, although multiple functions were identified through ABC charts.

Phase 2: Baseline and intervention.

Data was collected simultaneously for several participants during baseline (A) and return to baseline (A+; see Figure 3). One school supplemented behaviour recording with an electronic system due to staff time constraints. Behaviour recording was supplemented by two undergraduate research assistants and the primary researcher if required (completed for 10 sessions within two schools across A and A+). All sources of behaviour recording were combined for analysis.

Teaching staff wrote a social story for the target behaviour during baseline (A) and read it to participants during intervention (B; see Figure 3). Schools were already familiar with writing social stories, and manipulation was minimised. The researcher received a copy of the social story but was not involved in its development. Frequency of reading varied across schools; some students chose to read their own copy regularly, whilst others were unable to tolerate the social story on 10 occasions.

During A+, teachers opted whether to continue the intervention due to ethical concerns around withdrawal of a potentially beneficial intervention. As such, this period was labelled ‘A+’ as it cannot be considered a true baseline if the intervention continues. Where possible, participants completed the young person’s feedback form, with a completion rate of 37.5%, and two additional forms completed by educational staff (12.5%).

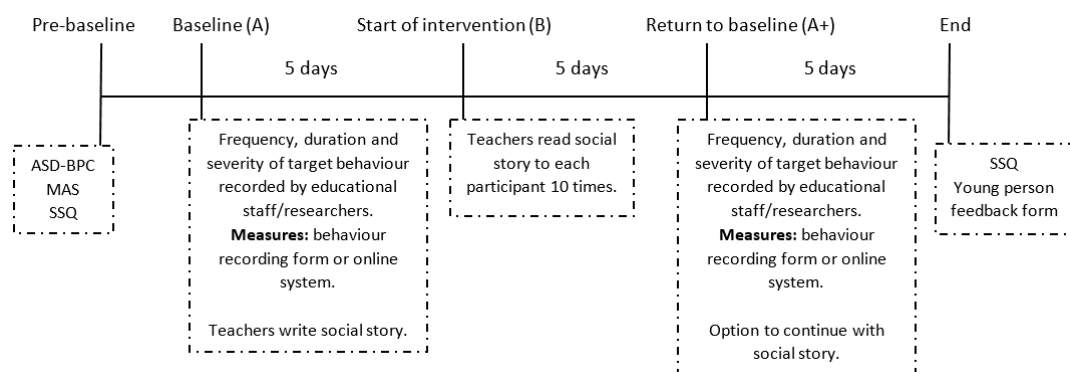


Figure 3. ABA+ study procedure including timescale and measures.

Statistical Analysis/Design (see Tables 9 and 10)

Table 9

Measurement of Study Variables

Variable	Measurement	Levels	Definition
Function	MAS and ABC charts	Four levels	Social attention Demand avoidance Tangibles Sensory
Independent variables	Coding system using data from functional assessment of behaviour at baseline and the intended outcome of coaching sentences within the story. Coaching sentences were assessed using ABC charts to determine the function which they were addressing. A second independent rater assessed function of the social stories using the same coding system to establish inter-rater reliability, with agreement of 88%. Discrepancies were resolved through discussion.	Two levels	Functionally congruent (FA+): match between function of behaviour and social story Not functionally-congruent (FA-): no match between function of behaviour and social story
Time	Stage of study process	Two levels	Pre intervention Post intervention
Treatment response	Assessed as change in measures of challenging behaviour from pre to post intervention	Two levels	Positive treatment response (TR+): decrease in behaviour of at least 25% Negative treatment response (TR-): decrease in behaviour of less than 25% or an increase in behaviour. Due to sample size, it was not possible to further distinguish between minimal/no change and increase in behaviour.
Dependent variable			
Social skills	Social Skills Questionnaire	Mean of scores pre-post	

Table 10

Analysis Methods for Hypotheses Testing

Hypothesis to be tested	Independent variable	Dependent variable	Analysis
Hypothesis one	Function	Treatment response	Fisher's exact test using contingency tables
			Odds ratio (effect size)
Hypotheses one to three	Functional congruence	Treatment response	Fisher's exact test using contingency tables
			Odds ratio (effect size)
Hypothesis four	Proportion of sentence types	Treatment response	Pearson's R correlation
Hypothesis five	Time	Social skills	Wilcoxon signed-ranks test

The pilot study was conducted in a naturalistic setting with no intervention manipulation, using an ABA+ case series design, where A was baseline, B was the social story intervention and A+ was return to baseline. A summary table for all participants can be found in Appendix C3. Data on frequency, duration and severity of behaviour during A and A+ was entered into a spreadsheet as total per session, with an overall total for A and A+ calculated across all episodes of behaviour per participant.

Completion rates of recording for duration and severity were considerably lower than frequency, with recording completed for 48% - 62% of total episodes. This was not deemed to be representative of total behaviour therefore further analysis was restricted to frequency. However, treatment response based on average duration and severity per episode of behaviour was calculated for informational purposes but not used in the main analyses (see Table 11).

Treatment response was calculated according to change in average behavioural frequency per recorded session from A to A+ (see Table 9 for a definition of positive and negative treatment response). Outcome of the intervention was calculated according to treatment response (n=15, one participant was excluded due to missing data during A+).

Participants were grouped by treatment response for further analysis (TR+ and TR-; see Table 12). Effect sizes were calculated to assess magnitude of change in frequency using the standardised mean difference (SMD) based on Glass' Δ calculation, with the denominator of standard deviation of A (Table 12).

Due to the small sample size, parameters for parametric testing were not met. As such, non-parametric independent group analyses (two-sided Fisher's exact test on a 2x2 contingency table) was used to examine the influence of the naturally occurring independent variable (IV) functional congruence on the dependent variable (DV) of treatment response in line with hypotheses one to three. Three participants were excluded from analysis; where function was uncodable in the social story (n=2) or due to missing data (n=1).

Additional analysis using a two-sided Fisher-Freeman-Halton's Exact test on a 2x3 contingency table was conducted into the association between function and treatment response (IV) as a precursor to functional congruence. Odds ratios were calculated for all contingency table analyses.

A non-parametric repeated measures analysis (Wilcoxon Signed Ranks test) examined change in social skills (DV) over time (IV). The SSQ was completed at both timepoints for 14 participants, with missing data at A+ for two participants. One item of data was missing pre and post for participant 001 (equating to 3.33% of all data); data was imputed using the replacement of the average method.

Sentence format was coded according to Gray's guidelines on proportion of sentence types (descriptive, coaching or violation of guidelines) and social story formula (number of descriptive sentences divided by coaching sentences = ≥ 2 ; see Table 2). A second rater independently coded the social stories; inter-rater reliability was 85% for the total number of sentences with identical coding. Discrepancies were resolved through discussion to reach 100% consistency. Four social stories (25%) adhered to the social story formula, with calculations across all social stories ranging from 0 – 3. However, all social stories which fulfilled the formula also contained violation sentences; therefore none were classified as fulfilling social story guidelines (Gray, 2015).

For analysis of the association between treatment response (DV) and presence of violating sentences within a social story (IV), participants were divided into two groups

according to inclusion of violation sentences, with one participant excluded due to missing data, and a two-sided Fisher's Exact test was used on a 2x2 contingency table.

In addition, post-hoc analysis using Pearson's R examined the association between magnitude of change (%) in frequency of behaviour and three variables independently: magnitude of change (%) on a social skills measure; proportion of sentence types and comprehension.

Table 11

Treatment Response (TR) for Frequency, Duration and Severity of Behaviour

Participant code	Average Frequency		TR frequency	Average Duration		TR duration	Average Severity		TR severity
	Pre	Post		Pre	Post		Pre	Post	
001	0.60	0.67	0	4.17	2.75	1	2.00	2.00	0
002	0.30	0.20	1	7.67	7.50	0	2.67	3.50	0
003	2.8	1.33	1	1.00	1.00	0	nc	nc	
004	5.5	nc		1.00	nc		nc	nc	
005	4.25	4.33	0	1.82	1.18	1	2.24	2.45	0
006	0.75	2	0	1.00	1.00	0	1.50	1.50	0
007	0.78	0	1	1.57	0.00	1	1.71	0.00	1
008	1.1	0.33	1	1.27	1.00	0	2.18	2.00	0
009	6.17	2.75	1	nc	nc		2.54	2.27	0
010	3.3	0.8	1	0.08	0.04	1	2.31	1.00	1
011	0.6	0.7	0	1.92	2.57	0	1.83	2.14	0
012	1.5	0.8	1	0.79	2.54	0	1.11	1.00	0
013	3.17	2.9	0	0.94	1.86	0	2.05	2.54	0
014	13.6	10.63	0	2.05	1.67	0	2.23	2.80	0
015	1	0.2	1	2.81	0.50	1	2.88	1.00	1
016	0.8	0.25	1	2.63	2.75	0	3.25	3.00	0

Table 12

Descriptive Statistics including Standardised Mean Difference (SMD) Effect Size within Treatment Response Categorisation

Treatment response (n=15)	Descriptive statistics					
	n (%)	Mean % change in frequency	Range of % change in frequency	SD of % change	Mean SMD	Range of SMD
Positive treatment response (TR+)	9 (60%)	64.72%	-33.33-100%	.20	.64	0.21-1.20
Negative treatment response (TR-)	6 (40%)	27.75%	-21.84-+166.67%	.69	.14	.03-1.30

Results

Treatment Response and Function

A two-sided Fisher-Freeman-Halton's Exact test on a 2x3 contingency table showed no significant association between treatment response and function of behaviour ($p=1.00$; see Table 13). Odds ratio (OR) calculations suggested that positive treatment response for behaviours with a sensory function was 0.75 times more likely than an escape function, and 1.13 times more likely than a tangible function. Positive treatment response was 1.5 times more likely for behaviours with an escape function than a tangible function. However, associations were non-significant (see Appendix C4 for graphs).

Functional Congruence

A two-sided Fisher's Exact test on a 2x2 contingency table ($n=13$; see Table 14); found no significant association between treatment response and functional congruence ($p=.685$). An odds ratio for positive treatment response demonstrated minimal difference between likelihood of positive treatment response when functionally congruent or non-congruent; challenging hypothesis two ($OR=1.11$).

Table 13

2x3 Contingency Table for Function and Treatment Response

Function	Treatment response		Total (%)
	TR+ (%)	TR- (%)	
Sensory	3 (20%)	2 (13%)	5 (33%)
Escape	2 (13%)	1 (7%)	3 (20%)
Tangible	4 (27%)	3 (20%)	7 (47%)
Total (%)	9 (60%)	6 (40%)	15

Table 14

2x2 Contingency Table for Functional Congruence and Treatment Response

Functional Congruence	Treatment response		Total (%)
	TR+ (%)	TR- (%)	
FA+	5 (38.5%)	3 (23%)	8 (61.5%)
FA-	3 (23%)	2 (15%)	5 (38%)
Total	8 (61.5%)	5 (38%)	13

Social Skills Questionnaire (SSQ)

No significant difference was found between SSQ scores pre and post intervention using Wilcoxon Signed Rank test ($Z=.824$, $p=.41$, $d=.16$; see Table 15), challenging hypothesis five.

Table 15

Outcome Pre and Post Intervention on the Social Skills Questionnaire (SSQ)

Pre-intervention		Post-intervention		Increase in SSQ scores			No change	Decrease in SSQ scores			
Range	Mean (SD)	Range	Mean (SD)	n (%)	Range of % increase	Mean (SD)	Positive TR (n)	n	n (%)	Range of % decrease	Mean (SD)
4-43	21.57 (9.20)	9-36	23.29 (8.36)	8 (57%)	4.5-125%	48.3% (.50)	4 (29%)	2 (14%)	4 (29%)	10.7-44%	15.7% (.17)

Social Story Structure

A two-sided Fisher's Exact test on a 2x2 contingency table ($n=15$; Table 16) to determine the association between treatment response and presence of violating sentences showed no significant association ($p=.486$).

Correlations

Pearson's R showed no significant correlations between magnitude of change (%) in frequency and magnitude of change on the SSQ ($r=-.334$; $p=.243$); proportion of

sentence types, even when proportion of violation sentences was high (see Appendix C5 for statistical results); or comprehension ($r=-.339$; $p=.217$).

Significant negative correlations were found between the proportion of violation and descriptive sentences ($r=-.801$, $p<.001$) and the proportion of violation and coaching sentences ($r=-.759$, $p=.001$); as violation sentences increased, descriptive and coaching sentences decreased. However, no significant association was found between the proportion of descriptive and coaching sentences ($r=.243$, $p=.382$).

Table 16

2x2 Contingency Table for Violating Social Story Sentences and Treatment Response

Contains violating sentences	Treatment response		Total (%)
	TR+ (%)	TR- (%)	
Yes	7 (47%)	6 (40%)	13 (87%)
No	2 (13%)	0	2 (13%)
Total	9 (60%)	6 (40%)	15

Case Series Approach

To further explore factors influencing treatment response in line with a case series approach, an in-depth comparison was drawn between the two participants with the largest decrease (007) and largest increase (006) in frequency of behaviour following the social story intervention (see Table 17); although conclusions are limited from two participants.

Young Person Feedback Form

Five out six children reported via the feedback form that they found the social story helpful and it would be useful to continue with it; of these, frequency of behaviour reduced for four participants (one had missing data). One participant reported not finding the social story helpful; although demonstrated a positive treatment response for frequency.

Table 17

Comparison between Participants at the Extreme Ends of the Range of Treatment Responses

Participant 006	Participant 007	Implications for hypotheses
% increase in frequency = 166.67%	% decrease in frequency = -100%	Highlights large variability in treatment response to a social story intervention
% decrease in SSQ = -44% (largest decrease out of 14)	% increase in SSQ = 82.4% (third largest increase)	Consistency between behavioural treatment response and change in social skills
Primary function = tangibles Secondary = attention	Primary function = tangibles	Identical primary function reinforces the finding of no association between function and treatment response
Functionally congruent	Non-congruent	Challenges hypotheses one to three that functional congruence leads to positive treatment response.
Fulfilled Gray's social story formula. Included violation sentences (n=1)	Fulfilled Gray's social story formula. Included violation sentences (n=2)	Raises questions about the influence of adherence to guidelines on effectiveness of a SS, although both stories included sentences which violated guidelines.
Comprehension level: second highest out of 16.	Comprehension level: within the bottom 25% of participants	challenging the influence of comprehension level on effectiveness.

Discussion

Overall, using an ABA+ intervention design, positive treatment response for frequency of challenging behaviour was found for nine out 15 participants (60%), with a decrease in frequency below 25% for two further participants; suggesting that the social story intervention was effective for the majority of the sample. However, hypotheses one, two and three around the role of function and functional congruence as moderators of treatment response were not supported. In addition, findings suggested that adherence to social story guidelines around structure was not associated with treatment response.

Finally, social skills were shown to improve for a small majority of participants (57%), tentatively lending support to hypothesis five although no significant difference was found between pre and post scores. Additionally, no association was found between magnitude of change on frequency of behaviour and social skills.

In relation to the first research question and hypothesis one, findings suggested that function did not influence treatment response; leading to the hypothesis that a social story intervention could be applied to a behaviour regardless of the underlying function. Attention was not rated as the primary function for any behaviour despite considerable association with challenging behaviour in the literature (for example a review by Hanley, Iwata and McCord, 2003, identified attention as the second most common function). However, it was identified as a secondary function for three behaviours; all of which were functionally congruent. Function was rated as escape and attention in the highest proportion of social stories, compared to tangible and sensory functions predominantly identified in the MAS. This is an interesting discrepancy given that both the MAS and writing of social stories was completed by teachers; suggesting that teachers' perceptions of behavioural function (and the required alternative behaviour) differed from ratings using a standardised measure. This hypothesis is partially supported by the proportion of non-functionally congruent social stories (38%). Based on literature showing higher effectiveness of interventions matched to function (Kokina & Kern, 2010; Pane et al., 2015); inaccurate perception of function might have implications for effectiveness of classroom interventions. However, findings of the present study did not support this hypothesis as treatment response remained high, and was not associated with functional congruence.

Several factors may have influenced this discrepant finding around functional congruence. Despite completing the MAS, teachers were not explicitly asked to write stories based on function, and attention was not drawn to functional congruence to minimise bias. As such, teachers may not have considered function when writing the social story, and functional congruence may have occurred by chance in some cases. In addition, reliability of a teacher-report measure (MAS) is limited compared to functional assessment involving manipulation of conditions. Although function was validated with ABC charts for a proportion of cases, multiple functions identified by ABC charts were not always picked up by the MAS, questioning precision of the measure. Findings may therefore have

differed with a different method of functional assessment, or if functional congruence had been manipulated as in Pane et al's study (2015); although this would have conflicted with the naturalistic study setting.

Addressing research question two and supporting hypothesis four, results on the SSQ suggested that social stories can influence social skills over a short period. This finding lends some support to Gray's proposal (1998, 2004) that social stories are effective due to improving social understanding, with implications for generalisable benefits beyond improving the target behaviour. However, correlational analysis demonstrated no association between improvement in social skills and behaviour. Furthermore, eight out of 14 participants showed a discrepancy between treatment response on behavioural frequency and social skills. This inconsistency may suggest that different mechanisms within social stories influence each variable, and further research could investigate relevant factors in effectiveness for social skills, challenging behaviour or both.

Results showed that none of the social stories fulfilled Gray's guidelines (2015) due to sentences that violated rules (n=4), a higher ratio of coaching to descriptive sentences than permitted (n=3) or both (n=9). Despite this, frequency of behaviour reduced for most participants including one social story consisting entirely of violation sentences, suggesting that sentence structure and adherence to Gray's guidelines (2015) may not be significant factors in treatment response for challenging behaviour. This supports previous research showing effectiveness of non-adhering social story interventions (Kokina & Kern, 2010; Quirnbach et al., 2009; Reynhout & Carter, 2006); and research showing low adherence rates to guidelines within educational settings (Reynhout & Carter, 2009).

As such, this pilot study significantly contributes to literature by challenging the influence of several hypothesised factors on treatment response to a social story. Factors including function, functional congruence, social story structure and comprehension were all non-significant in their association with treatment response, and inconsistencies were highlighted between behaviour and social skills. This raises the question: why were the social stories in the study effective?

A key consideration is the influence of environment factors on treatment response. Due to limited experimental control, other interventions may have occurred simultaneously during the intervention period. As such, the independent contribution of the social story

intervention cannot be disentangled from other interventions, highlighting a limitation of a naturalistic design.

Additionally, delivery of a social story involves other variables which could act as reinforcers for behaviour, such as provision of social attention; fulfilling the function of some behaviour and negating its necessity. Furthermore, study participation involved an increased focus on behaviour, which may have altered staff responses to behaviour due to hypervigilance and the introduction of a new intervention. As such, the setting context may have been altered during the study, thus indirectly contributing to changes in frequency. Other variables such as visuals within the social story may have contributed to reductions in behaviour independently of the written text, especially given low comprehension levels for most participants. This hypothesis is supported by the lack of association between sentence type and treatment response, raising questions about the role of story content on behavioural change.

Finally, Reynhout and Carter (2011a) suggested that social stories represent behavioural contingency contracts, producing positive outcomes through highlighting paired associations between stimuli and reinforcers. Research has shown behavioural approaches based on reinforcement schedules to be highly effective for challenging behaviour (Martinez et al., 2016). This hypothesis conflicts with Gray's proposition that a positive outcome occurs due to improvement in theory of mind and central coherence deficits (1998, 2004). A behavioural explanation is supported by the high proportion of violation and coaching sentences. Violation sentences often outlined direct associations between behaviour and a negative consequence, and coaching sentences outlined positive or negative reinforcement of an alternative behaviour. Alternatively, clear explicit instructions in relation to behaviour that a child can and cannot do (such as in violation sentences) may also support central coherence difficulties by drawing together relevant social information, in line with Gray's proposal (Frith & Happé, 1994; Gray, 2004).

Strengths and Limitations

This pilot study had several strengths and limitations in addition to those already outlined. A considerable strength was the naturalistic setting and use of educational staff as co-researchers. This increased ecological validity and provided valuable information about feasibility of research in school settings, which will inform design of the wider social

stories project (Professor Mark Brosnan and Dr Liz Smith), as well as supporting findings by Marshall et al. (2016) around suitable outcome measures and data collection within schools.

The naturalistic setting also highlighted limitations in generalising interventions tested in tightly controlled research studies, to real-world settings where implementation is variable. One example was adherence to social story guidelines (Gray, 2015); findings highlighted a possible training need within educational settings to improve consistency of the social story intervention.

Whilst a naturalistic setting was a strength for feasibility assessment, limitations included loss of experimental control for example over extraneous variables, resulting in reduced reliability of intervention delivery. This was compounded by limited researcher presence in schools to oversee quality and completeness of data collection, as well as practical barriers to reliability and feasibility such as school term times. Measurement of behaviour was relatively rudimentary; as such, treatment response was calculated based on restricted frequency counts in some cases. Limitations of methodology meant that complexities within analysis of challenging behaviour were not captured, for example changes in intensity or rate of behaviour as demonstrations of treatment response.

The limited success rate of recruitment highlights the challenges of research within schools, as only 33% of approached schools participated in the final sample. The number of pupils who participated represented a small proportion of those who were potentially eligible in each school, suggesting a barrier around access to participants. This may have represented a limitation in study design, with too high demands placed on schools in proportion to availability of the research team. As such, sample size was small, limiting generalisability of results and restricting analysis to non-parametric tests.

Conclusions

In conclusion, no significant associations were found between treatment response and variables including function and functional congruence, challenging the main study hypotheses. Tentative support was found for hypothesis five, suggesting the influence of social stories on social skills over a short time period, which has implications for using social stories beyond challenging behaviour. The study contributed several useful considerations around factors influencing effectiveness of a social story intervention.

Although functional assessment was not found to be a significant factor in this study, further research is needed to test reliability of the finding. Factors such as provision of attention through repeated reading of the social story, and increased awareness of behaviour through recording may have implications for clinical practice. The distinction between effectiveness for social skills and challenging behaviour would be a useful avenue for further research. In addition, future research should consider discrepancies between intervention guidelines and delivery in a naturalistic environment; as well as further exploring feasibility challenges which act as barrier to conducting high quality research into social stories.

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Executive Summary: The Relationship between Challenging Behaviour and Social Story™
Interventions: A Pilot Study in a Naturalistic Setting

May 2017

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Internal Supervisor: Dr Ailsa Russell

Aims and Background

The aim of the main research project was to investigate the link between how a social story intervention is written and implemented and the reasons behind challenging behaviour in children with ASD. Social stories are stories that describe a social situation or skill and how a child might respond best in that situation to develop social skills. They were developed according to specific guidelines that the written story must adhere to, including the type and number of sentences the story should include.

Research has shown that challenging behaviour has different purposes (or functions) that drive the behaviour. These typically fall into four categories: gaining social attention, escaping from difficult situations/unwanted demands, gaining access to a preferred activity/item, and sensory stimulation. The study aimed to investigate whether social story interventions were more or less effective depending on the function of a behaviour. Furthermore, it was hypothesised that if the reason behind behaviour was known, an effective social story could be written based on this to describe a more helpful way for the child to fulfil this purpose. In this way, the function of behaviour and the function addressed in the social story would match (labelled functional congruence in the study). By contrast, if the purpose is unknown then the social story may have less effect as it would not address the reason behind the behaviour (or would not match the function, labelled non-congruent).

In addition, research has shown that social stories can be helpful for two purposes: reducing challenging behaviour and improving social skills. As such, both outcomes were measured in the study. Finally, other variables that have been shown to influence the effectiveness of a social story intervention were investigated, such as comprehension, and adherence to social story guidelines.

Method

Data was collected through observations of challenging behaviour in a school setting by teaching staff, which were recorded using diary measures. Observations took place before and after the social story intervention was introduced. Social stories were written by teachers and read to children with ASD regularly. Teachers also completed a series of questionnaires.

Researchers analysed the social stories to see how well they responded to the function of the target challenging behaviour. Social stories were also coded to determine adherence to Gray's guidelines (2015). Data was analysed to investigate:

- Changes in frequency of challenging behaviour before and after the social story intervention (labelled treatment response)
- Changes in social skills pre and post intervention
- The influence of behavioural function on treatment response
- The influence of functional congruence (between the behaviour and social story) on treatment response
- The relevance of social story guidelines on treatment response
- The influence of comprehension on treatment response

Results

The majority of participants showed a reduction in the frequency of challenging behaviour after the social story intervention. This suggests that the intervention was largely effective. In addition, social skills improved after the intervention for a small majority of participants, supporting the hypothesis that social stories can be effective for social skills over a short period of time.

However, no significant associations were found between any variables, suggesting that function and functional congruence did not influence treatment response. Comprehension was also shown to have no association with outcome. Despite this, several interesting questions were raised as a result of non-significant findings. The findings challenged the hypothesis that tailoring an intervention to the underlying function of a behaviour resulted in a reduction in challenging behaviour. Findings also challenged the importance of Gray's social story guidelines (2015), since none of the social stories fulfilled social story criteria and many contained sentences which violated guidelines, however were still effective in reducing behaviour.

Findings also showed no association between improvement in challenging behaviour and improvement on social skills. This could suggest that there are different

mechanisms within social stories which are effective for social skills or challenging behaviour.

Conclusions and Implications for Clinical Practice

Findings challenged the importance of proposed factors that influence effectiveness in a social story intervention, such as matching interventions to function (functional congruence), adherence to social story guidelines and comprehension. This has implications for the way in which social stories are written in the future. Factors such as providing attention through reading the social story repeatedly may have implications for clinical practice, as well as other factors such as higher awareness of a behaviour which may have altered staff responses in the classroom.

One of the most important contributions of the study was around feasibility of conducting research in a naturalistic setting such as a school. The study highlighted challenges around recruitment, demonstrating the importance of ongoing researcher presence and a high level of contact with schools. Considerations around teachers as co-researchers included the need for brief and simple outcome measures due to time constraints. Discrepancies were found between real-world application of social story interventions and research findings, suggested a need to re-evaluate the effective mechanism within the intervention. The research also demonstrated the challenge of conducting high quality research within this field and highlighted areas for further research, such as investigating the independent contribution of factors within social stories to improving behaviour or social skills.

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Connecting Narrative

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Internal Supervisor: Dr Lorna Hogg

This connecting narrative aims to outline my experience and reflections on designing and conducting research and case studies over the course of training. I will reflect on the process including challenges and learning opportunities within each project in turn, including the case studies. I aim to conclude by drawing out similarities across my research experience during training as well as considering future aspirations for research as a qualified clinical psychologist. Prior to starting the course, my research experience had been relatively limited, although I had experience in audit and evaluation through the use of outcome measures in clinical settings. As such, designing and implementing several research projects throughout training provided an important learning experience which has equipped me for the challenges of balancing multiple demands as a qualified clinician, whilst also retaining the value of involvement in research.

Main Research Project

Development

Pre-training, I had a strong interest in working within the area of Autism Spectrum Disorder (ASD) through work and family experience, and entered the course keen to develop this through clinical practice and research. This became a strong theme throughout clinical placements, including working with individuals with ASD on both elective placements which offered the opportunity to develop my theoretical understanding and knowledge of intervention. When deciding upon an area of research, I was keen to explore interventions within autism, considering how research might be able to improve the efficacy and consistency of existing interventions, both in clinical settings and in schools. Having considered several possible topics within the population, my internal research supervisor Dr Ailsa Russell highlighted an ongoing project in the department relating to social stories. We met with the project team including my field supervisor, Dr Mark Brosnan, and agreed that my research could be a helpful pilot study focusing on one particular area of social stories, in order to inform feasibility of recruitment and development of methodology for the wider project. Due to recruiting in a non-NHS setting, it was not necessary to complete an IRAS ethics application; however, approval was obtained from the Psychology ethics board at the University.

Recruitment and Process

The practicalities of working within a school setting, and relying on teaching staff as co-researchers, presented a considerable challenge in this research project. Over the course of recruitment, only 5 of 15 schools that I approached about the research eventually took part. Even once a school agreed to participate, due to competing demands and time, schools only felt able to complete the process with 2-4 pupils, which represented a tiny proportion of the number of potentially eligible students on roll. Co-ordinating the limited time I had available for research, and available time within the stressful and highly busy school environment proved very difficult. In addition, practicalities such as fitting a three-week research process around school term-time significantly reduced the time periods when it was possible to begin the process with each school. Following limited success with the initial recruitment process, which involved the researcher meeting with parents of two schools at end of term events and gathering interest; the recruitment strategy was adapted to enable teaching staff to identify participants based on their knowledge about their pupils and who the research might be helpful for. Daily behavioural recording over two weeks for each child also presented challenges in consistency within and across schools. This involved finding a balance between fulfilling research requirements around methodological control, and feasibility of time that teachers could give to the research project. I was fortunate to have the support of two undergraduate psychology students offering a few days to go into schools as research assistants. They aided data collection through conducting behavioural recording, functional analysis validity checks and prompting teaching staff with data collection when e-mail reminders were not picked up. The final sample size was small ($n=16$) and the study was therefore underpowered, which was disappointing and frustrating given the number of hours that were put into recruitment and contacting/visiting schools (including one third of school visits to schools that eventually withdrew participation). However, this reflected the design of a three-week process for each individual participant.

Service User Involvement

Due to the project largely focusing on behavioural changes following a social story intervention, it also felt important to gather information about participants' personal experience of the social stories. I thoroughly enjoyed the experience of meeting with a group of young people with ASD to pilot a feedback form for participants to complete at

the end of the process. I wrote a social story for the class about research and providing feedback, and through meeting with each child and looking through the form, I gained some valuable insights into changes to make, for example the importance of layout. Alongside feedback from young people, methods of behaviour recording also developed during the process through feedback from teachers in the first school who took part. Teachers' reflections around the feasibility of recording behaviour whilst in the classroom led to a simplified layout, which may have been beneficial in increasing levels of data completion in subsequent schools. The feedback demonstrated the value of using expertise of professionals in the research setting when designing studies.

Reflections and Contributions

Conducting the main research project was a steep learning curve around the feasibility of research within naturalistic settings; as well as selling the importance and value of research to other professionals within a context that is epitomised by considerable pressures and demands of its own. The project contributed valuable insights for the wider social stories project into areas that were more successful as well as difficulties to consider when working in schools with young people with ASD. Examples included the importance of a researcher based within the setting, and the need for simple and brief outcome measures. I hope that these insights will also be useful when attempting to publish the research, and I intend to share the findings at the final year research conference. I experienced considerable frustration during the project that I was unable to provide support in schools on a more regular basis due to constraints on my time with other course demands. I learned to adjust my expectations of the quantity (and quality) of data that was needed in order to draw some helpful conclusions from the research. I also learned about feasibility of research designs, for example the use of an ABA+ design (baseline, intervention, return to baseline) led to a lengthy research process for each participant, thus reducing the sample size.

Service Improvement Project

Development

The idea for my service improvement project came about following a presentation by Dr Samantha Phillips and Dr Kirsty James, Clinical Psychologists in the local hospital-based Cystic Fibrosis team, during the first year of my doctorate. I became interested in the

challenges within cystic fibrosis (CF) around parenthood and pregnancy, and approached them to discuss this further. We worked together with my internal supervisor, Dr Catherine Butler, to design a project which built on previous research into sex and relationships within CF, particularly spending time developing a questionnaire which enabled data collection in a sensitive way, influenced by feedback from teams. We were aware throughout development of the possible ethical challenges that might arise when conducting research relating to sex. This included changing the target population of the project to adults with CF, rather than parents of paediatric patients due to concerns about considering sex and relationship issues in relation to young people.

Ethics

One of the most difficult parts of this project involved gaining approval for the project from the Research and Development (R&D) team. The project faced multiple setbacks, with an R&D process of around four months following approval from the Psychology board of ethics at the University. This included significant revisions to the project methodology and questionnaire, and considerable amounts of time meeting with members of the R&D team and clinicians in the Cystic Fibrosis team. The process of gaining R&D approval was supported by my internal and external supervisors through ongoing discussions and feedback on revisions in light of requirements from the R&D team. This was an important learning curve for me about the challenges of research in a 'taboo' area, and enabled me to develop skills in working with others in difficult situations and adaptability, whilst still advocating for the important elements of the research such as putting the patient's wellbeing as the centre of any decisions made during development.

Recruitment and Process

Recruitment using an online questionnaire enabled anonymity which was felt to be crucial when discussing a personal and sensitive topic. Despite this, the low response rate meant that other means of recruitment were necessary, and recruitment was greatly supported by an undergraduate psychology research assistant who attended several clinics and conducted visits to the ward to recruit more participants and pilot the questionnaire. Sharing results of the project with both adult and child cystic fibrosis teams was an interesting experience which highlighted some ongoing resistance to policy change around sex and relationships, despite results showing a clear need within services. However,

commitments were made around improvements to the procedure for sex and relationships with adolescents, and ongoing plans were made for consultation with parents and training for staff in the area which were encouraging outcomes.

Service User Involvement

This was an important part of the Service Improvement Project given the sensitive nature of the topic. During its development, the questionnaire was piloted on several occasions with patients on the respiratory ward who provided helpful feedback around methodology as well as the importance of the topic. Service user involvement throughout the project was a valuable reminder of the need to maintain a focus on improving services for patients where there were gaps in provision, even amongst frustration and changes in light of ethical procedures.

Reflections and Contributions

Overall, I greatly enjoyed the opportunity to gain knowledge in a specific area of health, and valued the numerous learning experiences provided over the course of the project such as presenting the research and background literature locally to teams, nationally and internationally. The frustrations of the R&D process also served to refine and improve the project through increased time spent on design and methodology, as well as enabling me to consider ways to facilitate research in taboo areas in my future career. Presenting at the national study day for clinical psychologists and social workers on the topic of taboos was an exciting experience, and outcome of the research was also shared via a poster presentation at the European conference in Switzerland, with the paper currently under review for publication in a peer-reviewed journal.

Literature Review

Development

The process of identifying a question for the literature review was lengthy, with several possible areas explored, and a change in direction from my original research proposal. I was keen to review research in an area that would be relevant to possible future career interests, for example within child and adolescent mental health. I also had an interest in anxiety, which has formed a thread throughout much of my clinical work and

case studies. My interest in selective mutism was raised due to reading in the area for possible clinical work during my CAMHS placement, and the associated links with both ASD and anxiety. Through discussion with my research supervisor, we constructed a research question to address a gap in the literature around cognitive factors in selective mutism. This area also enabled me to build on theoretical knowledge of cognitive models of anxiety acquired throughout training and awareness of development of new cognitive models through evidence-based practice.

Process

My experience with conducting literature reviews prior to training had been limited, therefore I was keen to develop skills in this area. The small amount of research on my chosen topic led to a need for creative thinking around ways to synthesise available literature. In order to produce a methodologically sound review, a systematic approach was taken to searching literature. However, the quality of identified studies was highly variable, with considerable reliance on case reports and qualitative data with informal methods of data collection. This presented a challenge in maintaining a rigorous approach to the synthesis as well as reducing bias. Considerable research into qualitative methods of synthesising literature led to agreement that an inductive content analysis approach would be best suited to address the research question. This provided a way of quantifying data whilst attempting to reduce bias and increase reliability. The presence of several competing theories around selective mutism led to interesting parallels drawn between findings of the review and previous propositions.

Reflections and Contributions

The review provides a basis for future development of a cognitive model in a similar way to other anxiety disorders as explored throughout clinical training and through case studies. It evaluated the current evidence for cognitions based on participants, and highlighted the importance of further research to build a strong evidence-base. In addition, the potential contribution of the review to developing an outcome measure aligns with the strong message of clinical psychologists as scientist-practitioners presented throughout training. This again highlighted to me the importance of having tools to measure clinical practice and outcome, in order to continue building evidence-based interventions in line with NICE guidelines.

Case Studies

Similarly to reflections on writing the literature review; the opportunity to write five case studies over the course of training was valuable in highlighting the role of research and building an evidence-base within clinical practice. My literature review relied heavily on case reports, and my main research project made use of a case series; both of which highlighted the value of insights that can be gained from a single case design. On reflection, I feel that my clinical practice was enhanced through the process of researching background literature for case studies and considering possible outcome measures. This was particularly relevant when using a single case experimental design, for example during my CAMHS and Learning Disability placements. The process provided an additional layer of depth to my clinical practice, encouraging me to plan the assessment and intervention through drawing on evidence-based cognitive models and using these within the formulation. The variety of clinical presentations throughout my case studies offered valuable learning opportunities to explore up-to-date research, and develop in-depth knowledge of several areas, from OCD to low mood, to selective eating.

However, the process also raised considerations around the discrepancy between clinical practice and research. For example, applying models and interventions developed through tightly controlled research trials to clinical settings according to NICE guidelines was challenging at times. This was particularly apparent when setting up collection of outcome measures for single case experimental designs, where at least three measurements were required before beginning intervention. This raised ethical challenges when clients were highly resistant to outcome measures or it was difficult to find appropriate measures that were short enough to repeat on a regular basis. Throughout training, I feel that I have refined my skills in presenting clinical work through case reports. I have broadened my awareness of the implications of each piece of work for clinical practice and improving clarity of the pathway to intervention based on a clear rationale founded in research.

Conclusions and Future Research

Overall, the experience of conducting research in a variety of ways across the course has been a helpful learning process. Challenges have arisen around ethical approval and practicalities of research in real-world settings. Equally, benefits have included developing skills in planning and implementing research studies and finding creative ways

to undertake reviews and adapt to circumstances. Each of the three research projects were targeted within my primary area of interest and intended career population of children and young people. This took place through direct work with children in the main research project, obtaining data to inform practice in the paediatric team within the service improvement project, and reviewing literature for a predominantly childhood disorder in the literature review. Another common thread across the projects and case studies was a focus on improving current knowledge and practice in order to benefit wellbeing. This included researching factors to improve efficacy of an intervention in the main project, evaluating and improving current service provision around sex and relationships education, and developing an understanding of an under-researched area within selective mutism in order to improve the applicability of interventions. The case studies were inherently focused on aiming to improve the wellbeing of the clients that I worked with, as well as increasing my own clinical knowledge and providing a case report with wider clinical implications,

I am keen to find ways to incorporate research into my career as a qualified psychologist, despite challenges with the pressures of increasing workload and diminishing resources in the NHS. The process of using outcome measures and writing up single case experimental designs is learning that I hope to apply and develop when qualified, as this presents an accessible way to continue contributing to an evidence-base and conducting research and evaluation even within a stretched NHS environment. In addition, I recognise the value of trainee clinical psychologists in providing opportunities to develop research interests through supervising projects and taking part in service improvement projects within teams; therefore this is something I would hope to engage with when qualified.

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'Don't worry about anything; instead, pray about everything. Tell God what you need, and thank him for all he has done. Then you will experience God's peace, which exceeds anything we can understand. His peace will guard your hearts and minds as you live in Christ Jesus.' - Philippians 4:6-7

Appendix A1. Codebook for Content Analysis

Main categories	Initial categories (sub)	Codes	Raw data	References
Beliefs about control over speaking	Lack of control	Not in their control	Being silent outside of their homes was not something they had decided or agreed upon. It just happened.	1
		Unable to break joint silence	They both confirmed that it would have been helpful if the other had started talking.	1
		Lack of control over speaking	She tells her parents she does not speak with other adults because "my brain wouldn't let me..."	3
		Lack of control over speaking	She said she "froze up" when talking to others and often felt like she could not respond despite wanting to, which she found distressing.	4
		Unable to speak	She would listen to what the other person was saying while focusing on her inability to speak.	4
		Lack of control over speaking	She experienced her symptoms as uncontrollable and cited that she would "freeze up" and be unable to respond.	4
		Lack of control over speaking	It wasn't something I wanted or something conscious, it just turned out that way. It was just like a huge mountain growing bigger and bigger. It was impossible to force that mountain. You can't suddenly start to speak when you haven't spoken for two years. (Catherine)	8
		Locked in silence	They felt relieved about being independent and no longer locking each other in a pattern of silence.	8
		Lack of control over speaking	Jakob told his mother when he was in 3rd or 4th grade: I thought that I would start to talk again. But it took such a long time and now I can't manage. Do you think I'm ever going to speak in school? (Jakob's mother).	9
		Lack of control over speaking	One of the recovered adults remembered feeling that it had been impossible to start talking when she had not talked for two years: It wasn't something I wanted or something conscious, it just turned out that way. It was just like a huge mountain growing bigger and bigger. It was impossible to force that mountain. You can't suddenly start to speak when you haven't spoken for two years (Catherine).	9
		Selective mutism in control	Hannah: It isn't me, I know who I am and I'm not shy or quiet, maybe that makes it harder. When I'm with my parents I can be myself but around everyone else it's like it [SM] takes over. I can get the words in my head but something won't let me say them and the harder I try the more of a failure I feel like when I can't.	15
		Unable to speak	It's as if something in my head stops working so it won't let me talk. Trying to talk is like a pressure building up and I start shaking until I feel like I'm going to cry and I just have to get out of there. Out of the situation. It's frustrating . . .	15
		Selective mutism in control	Sam: I know I can talk because there are people I do speak to but anywhere else it's like there's this subconscious roadblock that stops me saying anything.	15
		Selective mutism in control	Ben: I've always felt like I could talk if I could escape from this selective mutism.	15
		Lack of control	It never feels like anything I can control, even when all thoughts and efforts are directed towards speaking. Strangely the feeling that speaking is out of my own control extends to some situations where I do speak, which are still uncomfortable. Generally these are situations where speaking is required, such as asking for a train ticket. It's as if I were observing myself talking or being silent, both feel significantly different from	15

		situations where I feel as if I'm myself and can speak freely.	
	Prediction of being unable to speak	On one hand it helped, I wouldn't have been able to talk anyway.	15
	Lack of control	It can be hard with family, people who I care about and who care about me, but I don't get to be me or say the things I want to.	15
	Unable to function in the real world	Hannah: I don't think things will get better, if anything they'll get worse. My parents won't be here forever, I don't know what I'd do. How can I function in the real world when I can't even speak.	15
	Selective mutism in control	...without trying to fight off the feeling that at any moment selective mutism will kick in, and make me either stumble over my words or fail to say them at all.	15
	Attempts to control speech	He said, "This is how it happened. All of a sudden words just popped out of my mouth, even when I didn't want to say them. But there are still some words I don't say."	17
Active control	Active controlling efforts to speak	This often resulted in her abandoning efforts to speak	4
	Not talking else others would win	Elisabeth and Sarah felt that if they spoke, others would 'win'.	8
	Not talking else others would win	Elisabeth and Sarah felt that they could not start to speak, because if they did, then the others would 'win'.	8
	Not talking else others would win	Elisabeth and Sarah claimed that no person could ever have told them to start speaking, because they would never have allowed others to 'win'.	8
Beliefs about expectations of others	Determined not to speak	The adults who had recovered described themselves as having been absolutely determined not to speak.	9
	Refusal to speak	Indeed, the recovered adults said that under pressure to speak, the refusal to speak would get stronger.	9
	Chose not to speak	In the case of one participant who continued to meet criteria for selective mutism, she reported at follow-up that she simply chose not to speak in certain situations and was not fearful.	14
	Consciously withholding speech	She often greeted them by saying "I didn't speak at all today." When asked why, she frequently answered either, "Because I want to go to work with Mommy"	16
	Expectations of others	They both confirmed that it would have been OK to be talked to if they were not expected to answer back.	1
	Expectations of others	Both of them confirmed that it was helpful that they were talked to without being demanded an answer.	1
	Others don't expect us to speak	They could not talk there and others did not expect them to either: I think they just got used to or thought that, OK, that's how they are, and there's not much to do about it. They just accepted it, they gave up. (Elisabeth)	8
	Others expect us to speak	They did not feel comfortable about others expecting certain behaviours from them. Even today, if someone expected the women to speak in an unfamiliar situation, it could make them feel stressed and unable to talk.	8
	Expectations of others	The other respondents withdrew more and more too as they feared that they had to talk.	8
	Expectations of others	When no one had any special expectations of her communication, it was easier to start from scratch.	8

Factors related to anxiety	Fear of interactions involving speaking	Expectations of others	If they happened to say something or behaved in a new way, all the women found it difficult if people remarked on the change. People just had to behave as though everything was the same if they were to change their communication pattern.	8
		Prediction about expectations of others	It was difficult to communicate with other people. Therefore I knew that if there were strangers it'll be difficult to speak with that person, because now that person will shake my hand and ask my name (Elisabeth).	9
		Expectations of others	When I was at secondary school, because no one expected me to say anything it became kind of impossible to say anything, like, other kids just avoided me. Even the teachers would treat me differently. In History class the teacher would just skip past me when we had to read things out.	15
		Expectations of others	Hannah: Besides my parents I don't have anyone to talk to. It's a circle I can't escape from. I don't speak so no one speaks to me and then I'm left alone and there isn't anyone to talk to . . .	15
		Speaking is anxiety-provoking	She endorsed initially (via writing) that speaking during therapy sessions was extremely anxiety provoking for her.	4
		Unable to think due to anxiety	Ava reported that when she reached this point of 'paralyzing anxiety', her mind went blank and she would stop trying to talk.	4
		Speaking is anxiety-provoking	For example, on a Subjective Units of Distress Scale (SUDS) from 0 to 10, with 10 being the highest anxiety, she rated the task of reading words from a whiteboard to a therapist as a 7 and talking to therapist as a 9.	4
		Speaking is anxiety-provoking	Ava described that often when she began to speak, her mind went blank, her throat tightened, and she became very aware of her physiological symptoms.	4
		Speaking is anxiety-provoking	Given Ava's high level of anxiety initially speaking in sessions, the therapist and Ava created an early fear hierarchy related to speaking with the therapist (moving from lower-level items such as writing responses, to whispering words, to speaking at an increased volume).	4
		Speaking is anxiety-provoking	After she was able to speak in session, Ava, her mother, and the therapist created a hierarchy of additional social situations (e.g., answering questions when called on, reading aloud, asking for directions)	4
		Speaking is anxiety-provoking	Ava and the therapist created a new fear hierarchy specifically oriented to obtaining a summer job. This hierarchy included items including phone and in-person components such as calling a store to ask whether they were hiring, completing job interviews over the phone and in-person, greeting customers at a store, and talking with potential customers.	4
		Speaking is anxiety-provoking	The client reported the highest level of fear for all interactions that required speaking.	6
		Speaking is anxiety-provoking	For many patients, mutism was associated with remarkable anxiety states which were reported by 14 patients.	10

	Speaking is anxiety-provoking	Once a fear hierarchy was established in Session 5, the client and his mother rated Bruce's fear of each item each week on a scale from 0 to 10. Items on the fear hierarchy, in order of least to greatest anxiety, were (1) ordering a drink at a movie theater, (2) reading out loud with just the therapist, (3) answering a classmate's question, (4) talking to the school teacher, (5) checking in at the clinic, (6) talking to an unfamiliar adult in the therapy room, (7) asking a classmate a question, (8) reading out loud in front of the class, (9) talking to the school principal, and (10) reading out loud with the therapist and unfamiliar adults.	11
	Speaking is anxiety-provoking	During a two-week formal baseline, the therapist and student constructed a hierarchy of feared events related to speaking. Imagined speaking at school on weekend Spoke at school on weekend with mother and therapist Therapy session on school grounds during school hours Teacher, therapist, and student listen to audiotape of the student speaking Spoke one word to his favorite teacher Conversation with favorite teacher and therapist Spontaneous conversation with school counselor, therapist absent Conversation with teacher, peer, and therapist Spontaneous conversation with teachers, therapist absent Spontaneous conversation with coach, therapist absent Presentation of 5 minute videotaped speech to classmates Phone conversation with classmate SUD at baseline SUD in session Conversation with teacher, two peers, and therapist	12
	Speaking is anxiety-provoking	Furthermore, the student reported that he never spoke at school because it made him nervous...	12
	No longer fears speaking	The student denied feeling anxious when speaking at school	12
	Speaking is anxiety-provoking	But online I can be myself and say what I want without any of the stress and anxiety, even if I'm not really there with someone.	15
	Speaking is anxiety-provoking	Aaron: The efforts I've made so far to overcome SM have, I think, primarily been a fight against what would otherwise be a very bleak future. It's hard to imagine what it would be like to feel normal, to be in any situation and just speak without feeling anxious or uncomfortable	15
	Fear of speaking	Leah reports that she was..."afraid to talk," but not now.	16
	Negative social evaluation	"We stopped talking because they teased us about our dialect."	1
	Negative social evaluation	"And when we tried to say something, they sort of imitated us. And then they went on and said to everybody else that we said things in a certain way. And they said we were from southern Norway (which was not true)."	1
	Avoiding unwanted attention	She was highly selfconscious from an early age, and did not want others to focus attention on her.	8
	Negative evaluation	Sarah and Elisabeth thought that their behaviour had become more compulsive because their parents constantly criticized them.	8
	Fear of social evaluation	The recovered adults described themselves as highly self-conscious in their performance, and they felt that they had to take much responsibility for their own actions.	9
	Negative social evaluation	...he was worried about being rejected by his classmates.	12
Fear of evaluation	Fear of social evaluation	In addition, she indicated significant fear and avoidance of several evaluative stimuli, including answering questions in class, giving a report or reading aloud before the class, asking the teacher for help, conversing with others, and talking to people she did not know well.	13

Other fears	Fear of negative evaluation when unable to speak	Ben: At work I'd just hurry from place to place or try to look busy so no one would talk to me, you know, small talk? That way I could avoid looking like an idiot, sometimes someone would catch me and I'd try to mutter something at them. I think they gave up after a while.	15
	I will get it wrong	"I will say something wrong and won't be able to recover"	4
	Avoidance of getting it wrong	In addition, Ava was perfectionistic and reported that if she could not perform perfectly, she would avoid speaking entirely.	4
	Shyness	Jane also said that the reason she did not talk was because she was shy	7
	Fear of others	She was terrified by her authoritarian teachers and would say nothing to them in school. She said that she could not even think there, because of her anxiety.	8
	Shyness	This student was also faced with the unique challenge of answering questions from peers about his long-standing failure to speak. Thus, the therapist and the student collaborated to develop a list of possible responses. Response strategies that were generated included straightforward answers (e.g., "I am shy"; "I don't like to talk much").	12
	Worry about forgetting how to talk to people	Aaron: Having friends to speak to online was a huge help. It reassured me that despite having SM I could still communicate with people; I often worried that perhaps I'd forgotten how to talk to people.	15
	Shyness	Leah reports that she was "shy"	16
	Shyness	"...I was too shy."	16
	Role as the silent girl	None of the women could remember precisely when the mutism started, but they all remembered that they had gradually withdrawn more and more and developed a role as the silent girl.	8
Perception of separate identity as mute	Identity as non-speaker	Until then other people had defined her as mute, but when she spoke to people in the new country, she found that they reacted differently and expected her to talk: I didn't actually know who I was. I thought it was normal, that things are like this. It's only the huge inner anxiety that gave me a clue that this is not normal and I have to do something. The only way out was to travel away, but before that I thought that this is life. I didn't know anything else. (Maria)	8
	Girls who did not speak	Yet, the effect of bullying was not to encourage Elisabeth and Sarah to start talking, but rather to strengthen their need to remain in control and thus retain their separate identity as the girls who did not speak.	8
	Separate identity	They found it difficult to be like everyone else. It was embarrassing, but at the same time they wanted to be like the other pupils.	8
	Identity as non-speaker	Catherine felt that it was impossible to start talking after she had not talked for two years, because that would undermine her sense of self	8
	Them against us	The recovered adult twins had been determined not to speak, because if they did, others would "win" and they would lose their separate identity: We liked other people, but all the same, there were us two against the rest of the world. And we were not going to behave like them at all, and they were not going to be right (Sarah).	9
	Different but not excluded	Four reported that they had not felt socially anxious from an early age, even though they did not speak: "I joined everything, but I was quiet and different because I didn't speak. I was always told that I was quiet. Still, I never felt on the outside of the group (Hannah).	9

Beliefs about mutism as protective	To self	Positive consequence of not speaking	They felt acceptance and friendliness when other children helped them by talking on their behalf.	1
		Following rules	When asked why she didn't speak, Mimi wrote, "When I was little my mother told me don't talk to strangers."	5
		Self-protection	Linda felt that she had to make herself invisible and hide her capacities, because she was afraid of being laughed at by teachers who did not think that she actually had the knowledge she had.	8
		Self-protection	She described herself as trying to protect her personality, as nobody believed that she had the capacities she actually had.	8
		Self-protection	Maria locked herself in her own world and described her silence as a way of protecting herself against a chaotic world.	8
		Learned response to environment	The environment reinforced the girls' mutism in a variety of ways, and they learned that it was best to stay silent.	8
		Learned response to environment	One time when she happened to say something to the pupil next to her in the classroom, she was punished by the teacher and learned that it was best to say nothing.	8
		Negative consequences of speaking	Maria gave up trying to talk to adults after repeated disappointments.	8
		Others don't understand me	They tried to talk about something else, and she said that she felt misunderstood and disappointed, and gave up trying to talk to adults after this.	8
		Mistrust of others	Fourteen patients also reported a very sceptic attitude and mistrust of nearly all people in their environment.	10
		Failure to speak associated with conflict	The majority of the patients (n=17) also reported a connection of their symptomatology with conflict situations inside and outside the family.	10
	To others	Fear of negative consequences of speaking	Consequently, she kept silent in school because she feared she might inadvertently let slip out of her mouth the fact that she lived in a lesbian family.	2
		Speaking is dangerous	Tammy had taken quite seriously the adults' injunction not to tell anyone outside the family; she did not want to be responsible for bringing harm to her family.	2
		Fear of negative consequences of speaking	She said, "when Mom kept telling me not to tell nobody ever, ever, I know it was really, really, really bad!"	2
		Fear of negative consequences of speaking	Between sobs she revealed her fear that not only would Kaye and Barb be hurt, but she and her sister would have to live with their Dad if she inadvertently disclosed the facts about her home situation; she did not want to live with dad!	2
		Power of speaking (based on paper context)	One day his mother overheard a friend ask him, "Why don't you talk at school?" Jeremy replied, "Because I have a magic mouth."	17
Negative self-perception		I am a nobody	The boy expresses a feeling of humiliation and being a "nobody" even when he tries in vain to be acknowledged.	1
		Negative beliefs about voice	"...my voice sounds strange"	3
		Self-blame	This often resulted in her...engaging in self-blame.	4
		Mutism associated with shame and insufficiency	For many patients, mutism was associated with...feelings of shame and insufficiency (n=18).	10
		Shame and normality	The regret of silence may also merge into a shame of silence, an experience which Hannah describes: Hannah: The longer it goes on the more things I miss out on. Sometimes I feel ashamed because of that, and relying on my parents too much because it's not normal.	15
		Negative beliefs about normality	Lily: Talking to friends online helps a lot, that's when I can feel normal. Though sometimes it has the opposite	15

		effect too, someone might say something and I realise how not normal my whole situation is.	
Unknown reason	Unknown reason	Jane said she did not know why she was mute or enuretic.	7
	Unknown reason	Again, as with the previously described case studies, at the 7month follow-up with Chris, he had no idea why he did not speak in preschool or kindergarten.	7
	Unknown reason	Only 6 out of the 25 patients looked upon their mutistic reaction as a strange and unexplainable phenomenon.	10

Instructions for authors

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Aims and Scope. The *Journal of Clinical Child and Adolescent Psychology* (JCCAP) is the official journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53. It publishes original contributions on the following topics: (1) development and evaluation of assessment and intervention techniques for use with clinical child and adolescent populations; (2) development and maintenance of clinical child and adolescent problems; (3) cross-cultural and sociodemographic issues that have a clear bearing on clinical child and adolescent psychology theory, research, or practice; and (4) training and professional practice in clinical child and adolescent psychology as well as child advocacy. Manuscripts that discuss theoretical and/or methodological issues on topics pertinent to clinical child and adolescent psychology also are considered. Authors need not be members of Division 53 to submit articles to JCCAP.

There are several criteria that increase the likelihood that a manuscript will be favorably evaluated in JCCAP: (1) The paper reflects a substantive advance in our understanding of clinical child and adolescent psychology. (2) The paper is of such importance that it likely will influence an area of research. (3) The paper presents new ideas or creative methods. (4) The paper offers theoretically-driven hypotheses. (5) Multiple measures, informants, or procedures are used to collect data. (6) Sophisticated methodologies are carefully employed. (7) Longitudinal methods are used. (8) Data are rigorously and appropriately analyzed. (9) The implications of the findings for clinical child and adolescent psychology are well articulated.

Style of Manuscripts. Manuscripts should be prepared according to the guidelines in the *Publication Manual of the American Psychological Association* (6th edition; see www.apastyle.com). Typing instructions, including format, organization, and the preparation of figures, tables, and references appear

in the Manual. Manuscripts may be submitted as Regular Articles, Brief Reports, or Future Directions. A Regular Article may not exceed 11,000 words (i.e., 35 pages), including references, footnotes, figures, and tables. Brief Reports include empirical research that is soundly designed, but may be of specialized interest or narrow focus. Brief Reports may not be submitted in part or whole to another journal of general circulation. Brief Reports may not exceed 4,500 words for text and references. These limits do not include the title page, abstract, author note, footnotes, tables, and figures. Manuscripts that exceed these page limits and that are not prepared according to the guidelines in the Manual will be returned to authors without review. Future Directions submissions are written by leading scholars within the field. These articles provide a brief summary of important advances that are needed within a specific research or practice area pertinent to clinical child and adolescent psychology. Future Directions submissions are by invitation only and undergo peer review.

All Regular Article and Brief Report submissions must include a title of 15 words or less that identifies the developmental level of the study participants (e.g., children, adolescents, etc.). *JCCAP* uses a structured abstract format. For studies that report randomized clinical trials or meta-analyses, the abstract also must be consistent with the guidelines set forth by CONSORT or MARS, respectively. The Abstract should include up to 250 words, presented in paragraph form. The Abstract should be typed on a separate page (page 2 of the manuscript), and must include each of the following label sections: 1) Objective (i.e., a brief statement of the purpose of the study); 2) Method (i.e., a detailed summary of the participants, N, age, gender, ethnicity, as well as a summary of the study design, measures, and procedures; 3) Results (i.e., a detailed summary of the primary findings that clearly articulate comparison groups (if relevant); 4) Conclusions (i.e., a description of the research and clinical implications of the findings). Avoid abbreviations, diagrams, and reference to the text in the abstract. A list of up to five keywords that describe the central themes of the manuscript should be included below the abstract on page 2. *JCCAP* will scrutinize manuscripts for a clear theoretical framework that supports central study hypotheses.

In addition, a clear developmental rationale is required for the selection of participants at a specific age. The Journal is making diligent efforts to insure that there is an appropriately detailed description of the sample, including a) the population from which the sample was drawn; b) the number of participants; c) age, gender, ethnicity, and SES of participants; d) location of sample, including country and community type (rural/urban), e) sample identification/selection; f) how participants were contacted; g) incentives/rewards; h) parent consent/child assent procedures and rates; i) inclusion and exclusion criteria; j) attrition rate. The Discussion section should include a comment regarding the diversity and generality (or lack thereof) of the sample. The Measures section should include details regarding item content and scoring as well as evidence of reliability and validity in similar populations.

All manuscripts must include a discussion of the clinical significance of findings, both in terms of statistical reporting and in the discussion of the meaningfulness and clinical relevance of results. Manuscripts should a) report means and standard deviations for all variables, b) report effect sizes for analyses, and c) provide

confidence intervals wherever appropriate (e.g., on figures, in tables), particularly for effect sizes on primary study findings. In addition, when reporting the results of interventions, authors should include indicators of clinically significant change. Authors may use one of several approaches that have been recommended for capturing clinical significance, including (but not limited to) the reliable change index (i.e., whether the amount of change displayed by a treated individual is large enough to be meaningful, the extent to which dysfunctional individuals show movement to the functional distribution).

All manuscripts should conform to the criteria listed in Table 1 of the 2008 APA Publications and Communications Board Working Group on Journal Article Reporting Standards (published in *American Psychologist*). These reporting standards apply to all empirical papers. In addition, *JCCAP* requires that reports of randomized clinical trials conform to CONSORT reporting standards (<http://www.consort-statement.org/index.aspx?o=2965>), including the submission of a flow diagram and checklist. Nonrandomized clinical trials must conform to TREND criteria (see http://www.cdc.gov/trendstatement/docs/AJPH_Mar2004_Trendstatement.pdf) and meta-analyses should conform to MARS standards (see Table 4 in 2008 *American Psychologist* article).

Peer Review Process. *JCCAP* uses a two-tiered peer review process. All manuscripts are evaluated by the Editor or at least one Associate Editor to determine whether the manuscript is likely to make a significant impact to the scientific literature in clinical child and adolescent psychology. A significant proportion of manuscripts submitted to *JCCAP* are not selected for peer review, and this decision is typically made within 2–4 days after submission.

Manuscripts selected for peer review will undergo a masked review procedure. To prepare manuscripts for masked review, authors' names and affiliations should not appear on the title page or elsewhere in the manuscript file (they can be entered into the system and placed on a separate page in the cover letter file). Footnotes identifying the authors should be typed on a separate page and submitted in the cover letter file. Authors should make every effort to ensure that the manuscript file itself contains no clues to their identities. Manuscripts that do not comply with these instructions will be returned to the authors without review.

Publication Policies. Authors are responsible for all statements made in their work and for obtaining permission from copyright owners to use a lengthy quotation (500 words or more) or to reprint or adapt a table or figure published elsewhere. Authors should write to both author(s) and publisher of such material to request nonexclusive world rights in all language for use in print and nonprint forms of the article and in future editions. This applies to direct reproduction as well as "derivative reproduction" (where you have created a new figure or table which derives substantially from a copyrighted source). Authors are required to sign an agreement for the transfer of copyright to the publisher. All accepted manuscripts, artwork, and photographs become the property of the publisher.

Submitting Manuscripts. *JCCAP* receives all manuscript submissions

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Search Engine Optimization. Search Engine Optimization (SEO) is a means of making your article more visible to anyone who might be looking for it. Please consult our guide [here](#).

Appendix B1. Relationships and Sexual and Reproductive Health in Cystic Fibrosis

Questionnaire

Section 1: The role of hospital-based paediatric CF services in providing relationship, sexual and reproductive health information / support

We would be interested to hear your views about the role of paediatric CF teams in providing information and support to young people with CF. *(Please note that this information would always be provided at an appropriate age, depending on each patient's circumstances and needs).*

1. Please tick the boxes to show what extent you agree or disagree with the following statements:

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
Paediatric CF teams should provide patients with CF-specific advice about sexual and reproductive health.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	
Paediatric CF teams should provide patients with general information, advice and support about sexual and reproductive health.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	
Paediatric CF teams should provide regular updates / refreshers to their patients about sexual and reproductive health.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	
The CF team should not provide sex and relationships education, unless a patient specifically asks for this.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	
The CF team has a role to play in supporting patients to discuss sex and relationship issues with their prospective sexual partners.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	

- a. Any comments about your answers to Q1:

Section 2: Your own experiences of receiving sex and relationships information / support as a paediatric CF patient

This section of the survey is about your own experiences of receiving information and support from your hospital-based CF service as a paediatric patient (up to age 18).

2. Overall, how useful did you find the sex and relationships education/information and support provided by:

	Very useful	Fairly useful	Not very useful	Don't know/can't remember
Your school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Your parents	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Your hospital paediatric CF service	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- a. Any comments about your answers to Q2:

3. If there were any sources of information about sex and relationships that you found particularly useful as a paediatric patient, please write these below.

4. When you were a paediatric CF patient, did you **feel able** to raise sexual and reproductive health related issues with a member of your hospital CF team?

Yes ☐

No ☐

Can't recall ☐

5. When you were in the paediatric CF service, **how likely** were you to seek information/advice/support about sexual and reproductive health from a member of the hospital CF team?

Very likely	Fairly likely	Fairly unlikely	Very unlikely	Don't know/can't remember
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- a. If you answered “fairly unlikely” or “very unlikely”, what were the reasons for this?

6. To what extent did the paediatric CF service meet your information / support needs relating to the following topics? Please tick the relevant box for each topic.

	I got all the information / support I needed	I would have liked more information / support	I didn't want this information / support	Not sure / can't remember
Contraception	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
IVF	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Pregnancy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Fertility	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Parenthood	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. Is there any other information/support specific to sexual and reproductive health and relationships that you didn't receive but would have been useful for the paediatric CF service to provide to you?

8. Was there any information about sexual and reproductive health/relationships provided by the paediatric CF team that you found particularly helpful?

9. Is there anything you think **could have been done better** in providing you with information/support about sexual and reproductive health related issues?

Section 3: Information and support from the adult CF service

This section asks about your current experience of information and support around sexual and reproductive health and relationships from the **adult** CF service at the Bristol Royal Infirmary.

Have you ever sought information, support or advice about sexual and reproductive health and relationships from the **adult** CF service?

Yes ☐

No ☐

Not sure ☐

10. If no, what are the reasons why you haven't sought this information from the adult CF team? (*Tick all that apply*).

I haven't needed this information ☐

I don't feel I can ask the team ☐

I prefer other sources of information ☐

Other reason (please state):

11. If yes, what advice / support / information did you seek (*tick all that apply*):

Contraception ☐

Assisted reproduction e.g. IVF ☐

Fertility ☐

Pregnancy ☐

Parenthood ☐

12. Was your request for this information:

a. Fully met ☐

b. Partially met ☐

c. Not met ☐

If your request was not fully met, please comment why:

13. Any other comments/suggestions about the provision of sex and relationships education/support in the adult CF service:

--

Overall Evaluation

Overall, how would you rate the information, advice and support that you have received as a CF patient in relation to sex and relationships?

Excellent	Good	Fair	Poor
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Any other comments:

--

We are hoping to carry out follow-up interviews about this topic later in the year, and will be contacting adult patients within the Bristol CF service with further information soon.

If you are keen to take part in an interview and would like to be contacted directly, please put your e-mail address in the box below (we will detach this information from your responses to the questionnaire):

--

Demographic information

1. Gender:

- a. Male ☐
- b. Female ☐
- c. Prefer not to say ☐

2. Age group (please tick the relevant box):

- | | | | |
|-----------|--------------------------|-----------|--------------------------|
| 18-24 yrs | <input type="checkbox"/> | 25-34 yrs | <input type="checkbox"/> |
| 35-44 yrs | <input type="checkbox"/> | 45-54 yrs | <input type="checkbox"/> |
| 55-64 yrs | <input type="checkbox"/> | 65+ yrs | <input type="checkbox"/> |

3. Which of the following best describes how you think of yourself?

- a. Heterosexual / straight
- b. Gay / lesbian
- c. Bisexual
- d. Other
- e. I would prefer not to say

4. Marital status:

- a. Single ☐
- b. Married ☐
- c. Cohabiting ☐
- d. Separated/Divorced ☐
- e. Widowed ☐

5. Do you have any children?

a. Yes ☐

b. No ☐

a. If yes, how old are your children? (Please tick all boxes that apply).

0-4 yrs ☐ 10-14 years ☐ 19+ years ☐

5-9 yrs ☐ 15-18 years ☐

6. Age of diagnosis with Cystic Fibrosis: ____ years

7. At what age did you start attending the Paediatric Cystic Fibrosis service at Bristol Royal Hospital for Children?

a. ____ years

b. N/A – I have never been open to the Paediatric Cystic Fibrosis service at Bristol Royal Hospital for Children.

To complete the survey and submit your answers, please click 'Finish'. You will be unable to edit your responses after this point.

By clicking 'Finish', you are consenting to your data being included as part of the service improvement project, including analysis and reporting of results. All data will be anonymised and you will not be identifiable from the results.

Appendix B2. Confirmation of Ethical Approval including Amendments by the University of Bath Psychology Ethics Committee

psychology-ethics | Emily Norris 07/01/2016

RE: Ethics Reference Number 15-243 Emily Norris

Flag for follow up. Start by 08 January 2016. Due by 08 January 2016.

MessageHeaderAnalyzer + Get more add-ins

Dear Emily Norris

Reference Number 15-243: Relationships and sexual and reproductive health in individuals with cystic fibrosis: A service-directed approach

Thank you for satisfactorily attending to those amendments by providing this additional clarification. I can now confirm that you have full ethical approval for your study.

Best wishes with your research,

Dr Michael J Proulx
Chair, Psychology Research Ethics Committee

psychology-ethics | Emily Norris 26/05/2016

Re: Ethics 15-243

You replied to this message on 13/06/2016 19:31.
This message is part of a tracked conversation. Click here to find all related messages or to open the original flagged message.

MessageHeaderAnalyzer + Get more add-ins

Dear Emily,

Thank you for submitting these changes. The amendments are approved via Chair's Action.

Best wishes,
Michael

From: Emily Norris
Sent: Monday, May 23, 2016 8:33 PM
To: psychology-ethics
Subject: Re: Ethics 15-243
Dear Dr Proulx,

Reference Number 15-243: Relationships and sexual and reproductive health in individuals with cystic fibrosis: A service-directed approach

Thank you for confirming the process for submitting amendments based on R&D changes to my service improvement project since the initial submission and approval. I have now received approval from the questionnaire department of R&D (QJS) at UHBristol (see attached e-mail). I have attached the revised CF questionnaire and ethics attachments (information sheet, consent form and debriefing form). I have listed a summary of key changes below. I have also put these changes in the ethics application form as I was not sure if this was needed too (see attached).

1. Participants have now been given the option of completing the survey in person or via telephone, as well as online.
2. The survey has been considerably shortened and edited to meet QJS requirements, due to concerns about asking lots of open questions around a sensitive topic such as sex and relationships education. A second phase of follow-up interviews has been introduced to capture the data lost from the original questionnaire. The interview schedule will be constructed separately to the survey partially based on responses, and I will submit amendments to this phase of the study once the schedule has been completed. Consent will be obtained separately for the interviews.
3. Participants are given the option of including an e-mail address with their survey if they are keen to take part in the follow-up interviews - this data will be removed from the questionnaire responses and stored separately in a password-protected file before data analysis. As such, data will remain anonymised. This has been explained to participants on the advert, information sheet and debriefing form.
4. Participants have been made aware that they will be unable to withdraw from the study once they submit their responses to the questionnaire as these will be anonymous so it will not be possible to identify them. They can still withdraw before or during the survey.

Please do let me know if you require any further information.

Best wishes,

Emily Norris
Clinical Psychologist in Training

Dear Emily,

Thank you for submitting this request. Your amendments are approved via Chair's Action.

Best wishes,
Michael Proulx

From: Emily Norris
Sent: Tuesday, June 28, 2016 8:07 PM
To: psychology-ethics
Subject: Reference Number 15-243

Dear Dr Proulx,

Reference Number 15-243: Relationships and sexual and reproductive health in individuals with cystic fibrosis:
A service-directed approach

I am submitting a request for an amendment to chair's approval please - I have recently published my online survey and sent out the link via e-mail to patients, however have since realised that not all patients have an e-mail address with the team, therefore some will need a questionnaire to be posted. I am requesting permission to add this as a distribution method for the questionnaire please?

The advert, information sheet and debrief form attached are the same but have had the words tweaked to refer to both the option of completing the survey online or as a paper survey. The consent form contains identical information but the format has been changed to match a conventional paper consent form requiring a signature. The paper questionnaire has already received ethical approval as I was planning on handing this out at clinic which I have permission for already.

Best wishes,

Emily Norris
Clinical Psychologist in Training

Appendix B3. Ethical Approval from UHBristol Research and Development QIS Department

Lewis, Paul <Paul.Lewis@UHBristol.nhs.uk> Thu 19/05/2016 16:53

To: Emily Norris; Cc: Phillips, Samantha <Samantha.Phillips2@UHBristol.nhs.uk>; James, Kirsty <Kirsty.James@UHBristol.nhs.uk>; Catherine Butler;

Dear Emily,

Thank you for your updated questionnaire - you have QIS approval for this survey now. There are a couple of very minor changes to the questionnaire in the attached – a slightly truncated first paragraph (there were a lot of “ands” in one of the sentences in the last version!), and you need to use the correct name for the hospital. I’ve highlighted these in green - unless you have any major objections can you just run with these. We also wondered whether you should ask a demographic question around sexual orientation: given the content of the questionnaire this might be relevant. It’s up to you whether you put this in – but if you do, can you please use a recognised version of this question (I can’t recall if there is one in the Census, if not the attached Q35 is the one that we and the national surveys use, so that might help).

Collecting an email address within the survey potentially compromises anonymity. We don’t have objections to this as long as 1) you permanently separate the email address from the response data before any analysis starts, 2) you carefully follow IG Policies around the collection and storage of personal information, 3) you inform participants about this (mainly in the PIS, but also a note about it in the actual invite), and briefly make it clear what will happen to this personal info. Up to you how you proceed with this - if you need any advice let me know.

Fine to use the PIS / debriefing form if this is a requirement. There are a few bits and pieces in relation to this (again I’ve highlighted these in green in the attached):

- You may want to check that the aims of this questionnaire are still described accurately (fine if so)
- I wasn’t sure what “at the age range” means so could you tweak this
- You may want to revisit whether it will take 15-30 minutes, it might actually be less now
- Our R&I folks get a bit jumpy when service evaluation projects are referred to as “research”, so could you tweak this to something like “service evaluation” or “project”
- I wasn’t sure that the statement about being able to withdraw from the study at any time was correct. If the questionnaire is completed anonymously, how will you be able to withdraw that data?
- The line about the questionnaire not using deception was a bit odd and it doesn’t send a great message to our patients (i.e. that we’d potential deceive them) – please can you take this out

I’ll leave these for you to address (I don’t need to see it again from here unless you want me to).

I know you are anxious to get going with this, you did very well to start this project in good time. It’s important though to recognise that the last four months haven’t been “delays” or an “R&D process” - it’s been research design: getting a self-completed questionnaire that is fit for purpose, ensuring that the right methodology is adopted to generate high quality data for both your project and our service, and ensuring that we can provide a positive environment for you to conduct this sensitive research in (particularly as the service has had some challenges recently). This is time well spent as a researcher. So the length of time this has taken has, in the main, been a reflection of how much time we’ve had to invest on our side to get these things right (which we’ve been more than happy to do of course).

Best of luck with the survey, please don’t hesitate to get in touch if needed as it progresses. I look forward to seeing the interview schedule in due course.

Regards, Paul
Paul Lewis
Patient Experience Programme Manager
University Hospitals Bristol NHS Foundation Trust
Trust HQ
Marlborough Street
Bristol
BS1 3NU
Tel: 0117 342 3638

Appendix B4. Ethics Documents for Participants

Relationships and Sexual and Reproductive Health in Cystic Fibrosis



If you choose to take part, you will be asked to complete a short online survey. Survey responses will be anonymised by removing any identifying information.

The survey asks questions about:

- ❖ Your experiences of sex and relationships education in different settings
- ❖ How the Cystic Fibrosis team should provide information on relationships and sexual and reproductive health relevant to CF

The information you give us will directly affect changes within the CF service in Bristol.

If you would like to take part in the online survey, either:

- ❖ Type the web address below into your browser to begin the survey
- ❖ OR follow the web link in this e-mail

If you would prefer to complete the survey in a face-to-face or telephone interview (both will be audio-recorded), please contact the researcher using the details below.

Survey link: <https://bathreq.onlinesurveys.ac.uk/relationships-and-sexual-and-reproductive-health-in-cystic> **The survey password is:** [bristol](#) (all lower case)

We look forward to hearing your views!

Any questions? Please contact:

Primary researcher: Emily Norris, Clinical Psychologist in Training: en325@bath.ac.uk
OR Dr Samantha Phillips/Dr Kirsty James, Clinical Psychologists: 0117 3428168

Relationships and Sexual and Reproductive Health in Cystic Fibrosis: A Service-Directed Approach

Participant Information Sheet and Statement of Consent Online V.3

You are invited to take part in the following service improvement project. Please read the following information about the project carefully. You will be asked for your consent to take part before the questionnaire begins on the next page. If you have any questions, please contact the researchers using the details below.

Why is this study being done?

We are interested to know whether you received enough information about relationships and sexual and reproductive health when you were growing up, particularly from the Cystic Fibrosis service. We are hoping to use this information to make improvements to the way in which the Bristol Cystic Fibrosis service provides information on this topic to patients.

What will it involve?

You will be asked to complete an online questionnaire asking about your experiences of receiving information about relationships and sexual and reproductive health. The questionnaire is expected to take around 10-20 minutes and can be completed online. If you would prefer to complete the questionnaire in a face-to-face discussion or over the phone, this can also be arranged by contacting the researchers using the details below.

A few weeks after completing the questionnaire, you may be invited to take part in a follow-up interview. There is no obligation to do this and consent will be gained separately.

Confidentiality

All information collected about you during the course of the project will be kept confidential and will conform to the Data Protection Act of 1998. All paper-based and electronic information will be locked and password protected with access restricted to study personnel. Identifying data such as e-mail addresses will be separated from questionnaire responses before analysis and access to this information will be password-protected. As such, information will be anonymised so you cannot be identified.

We hope to report our findings in academic/health related journals and present them to relevant health professionals at meetings and conferences. You will not be identified in any reports or publications arising from the study.

Possible advantages

Information collected from you and other participants may help to improve our understanding of sex and relationships education within cystic fibrosis. Results of the project will be discussed with the Bristol Cystic Fibrosis team and will inform service provision in this area in the future.

Possible disadvantages

We don't consider there to be any disadvantages to taking part. The topic of relationships and sexual and reproductive health can be sensitive to discuss for some people, and may bring up questions or uncomfortable emotions. If you feel upset or distressed by any of the

research, you are invited to contact one of the Clinical Psychologists from the Cystic Fibrosis team who will offer you the opportunity to speak about this (contact details below).

What if there is a problem?

If you have any concerns or wish to complain about any aspect of the way you have been approached or treated as part of this study, you should initially contact the researchers who will do their best to answer your questions. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure (details can be obtained from your Primary Care/NHSTrust) or you can contact the University of Bath Psychology Ethics Committee at psychology-ethics@bath.ac.uk.

Consent and Right to Withdraw

If you would like to participate in the questionnaire, please respond to the consent statement below and click 'Next' – the questionnaire will begin on the following page. There is no obligation to take part in the study. If you decide to take part and then later change your mind, you can withdraw without giving your reasons either before you start or during the study. However, as all questionnaire responses are anonymised, you will be unable to withdraw your participation once you have submitted the completed survey. Taking part, or otherwise, in the study will not affect the treatment that you are currently receiving or are likely to receive in the future.

Researcher Contact Details:

Emily Norris (Primary researcher)
Clinical Psychologist in Training
(University of Bath)
Email: emily.norris@bath.ac.uk

Dr Samantha Phillips / Dr Kirsty James
Clinical Psychologists in the Cystic Fibrosis service
Phone: 0117 3428168

Dr Catherine Butler
Clinical Psychologist
Research Supervisor at University of Bath
C.A.Butler@bath.ac.uk

Statement of Consent for the Questionnaire

I have read and understood the information about the questionnaire phase of this study. I understand I am free to withdraw from this study before or during the survey without giving a reason for withdrawing, but will be unable to withdraw my responses once submitted as they will be anonymised. I understand that I can omit questions on the questionnaire that I do not wish to answer. In consenting, I understand that my legal rights are not affected. I also understand that data collected as part of this research will be kept confidential and that published results will maintain that confidentiality. Finally, I understand that if I have any questions about my rights as a participant in this research, or if I feel that I have been placed at risk, I may contact the chair of the Ethics Committee, Psychology, University of Bath, BA2 7AY, UK. E-mail: psychology-ethics@bath.ac.uk. I certify that I am 16 years or older. I have read the above consent statement and I give consent to participate in the above described study. ○

Consent Form Paper Questionnaire

- I have read and understood the information about the questionnaire phase of this study.
- I understand I am free to withdraw from this study before or during the survey without giving a reason for withdrawing, but will be unable to withdraw my responses once submitted as they will be anonymised.
- I understand that I can omit questions on the questionnaire that I do not wish to answer.
- In consenting, I understand that my legal rights are not affected. I also understand that data collected as part of this research will be kept confidential and that published results will maintain that confidentiality.
- Finally, I understand that if I have any questions about my rights as a participant in this research, or if I feel that I have been placed at risk, I may contact the chair of the Ethics Committee, Psychology, University of Bath, BA2 7AY, UK. E-mail: psychology-ethics@bath.ac.uk.

I certify that I am 16 years or older. I have read the above consent statement and I give consent to participate in the above described study (please circle):

Yes / No

Signed

Date

Name in Block Letters:

Signature of researcher

Date

Relationships and Sexual and Reproductive Health in Cystic Fibrosis: A Service-Directed Approach

Debriefing Statement V.2

Thank you for completing the survey. All of the data collected in this survey will be anonymised by removing any personal identifying information such as e-mail addresses and storing this separately to the data in a password-protected file. Therefore, if you do not wish to receive any further e-mail prompts to complete this survey or follow-up interview, please send this request via e-mail to: CFadults@UHBristol.nhs.uk with the subject line 'SRH Research Study'.

The aim of this service improvement project was to learn more about experiences of sex and relationships education amongst individuals with cystic fibrosis. In particular, we were interested in how the Cystic Fibrosis service shares information about sexual and reproductive health and relationships with patients and whether this fits with how patients would like to receive this information. We were also hoping to find out at what age patients wanted to receive this information from the Cystic Fibrosis team. Your data will help to shape how the Cystic Fibrosis Service in Bristol provides sex and relationships education in the future.

Once again results of this study will not include your name or any other identifying characteristics. You may have a printed copy of this summary if you wish, and you may also request a summary of the research findings once the project is completed if you wish. Some of the topics asked about in the survey may bring up uncomfortable or difficult emotions for some people. If you feel upset or distressed by any of the research, you are invited to contact Dr Samantha Phillips or Dr Kirsty James, Clinical Psychologists in the Cystic Fibrosis team, on 0117 3428168 who will offer you the opportunity to speak about this. If you have any questions about the research, please contact the primary researcher Emily Norris on en325@bath.ac.uk or Dr Catherine Butler on C.A.Butler@bath.ac.uk.

For further information on relationships or sexual and reproductive health in CF, please visit the following website, which was created by a group of professionals working with individuals with Cystic Fibrosis across the UK: <http://www.cfinfo.org/>

Thank you for your participation in this project.

You have now completed the survey and can close your browser.

Instructions for Authors

EDITORIAL PROCEDURE

Double-blind peer review

This journal follows a double-blind reviewing procedure. Authors are therefore requested to submit:

- A blinded manuscript without any author names and affiliations in the text or on the title page. Self-identifying citations and references in the article text should be avoided.
- A separate title page, containing title, all author names, affiliations, and the contact information of the corresponding author. Any acknowledgements, disclosures, or funding information should also be included on this page.

MANUSCRIPT SUBMISSION

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

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Online Submission

Please follow the hyperlink “Submit online” on the right and upload all of your manuscript files following the instructions given on the screen.

GENERAL

Inquiries regarding journal policy, suitability of the paper for the journal, and other such general topics should be sent to Editor-in-Chief Hough at: sigmund_hough@hms.harvard.edu

TITLE PAGE

Title Page

The title page should include:

- The name(s) of the author(s)
- A concise and informative title
- The affiliation(s) and address(es) of the author(s)
- The e-mail address, and telephone number(s) of the corresponding author
- If available, the 16-digit ORCID of the author(s)

Abstract

Please provide an abstract of 150 to 250 words. The abstract should not contain any undefined abbreviations or unspecified references.

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

GENERAL MANUSCRIPT GUIDELINES

All manuscripts should be in English. All manuscript pages (including figure-caption list, tables, and References list) should be double-spaced and use generous margins on all sides. Manuscripts should be checked for content and style (correct spelling, punctuation, and grammar; accuracy and consistency in the citation of figures, tables, and references; stylistic uniformity of entries in the References section; etc.). Empirical articles should include standard sections, such as Introduction, Methods, Results, and Discussion.

TEXT

Text Formatting

Manuscripts should be submitted in Word.

- Use a normal, plain font (e.g., 10-point Times Roman) for text.
- Use italics for emphasis.
- Use the automatic page numbering function to number the pages.
- Do not use field functions.
- Use tab stops or other commands for indents, not the space bar.
- Use the table function, not spreadsheets, to make tables.
- Use the equation editor or MathType for equations.
- Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables. Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

REFERENCES

Citation

Reference citations in the text should be identified by numbers in square brackets. Some examples:

1. Negotiation research spans many disciplines [3].
2. This result was later contradicted by Becker and Seligman [5].
3. This effect has been widely studied [1-3, 7].

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text. Do not use footnotes or endnotes as a substitute for a reference list.

The entries in the list should be numbered consecutively.

- Journal article
Hamburger, C.: Quasimonotonicity, regularity and duality for nonlinear systems of partial differential equations. *Ann. Mat. Pura Appl.* 169, 321–354 (1995)
- Article by DOI
Sajti, C.L., Georgio, S., Khodorkovsky, V., Marine, W.: New nanohybrid materials for biophotonics. *Appl. Phys. A* (2007). doi:10.1007/s00339-007-4137-z
- Book
Geddes, K.O., Czapor, S.R., Labahn, G.: *Algorithms for Computer Algebra*. Kluwer, Boston (1992)
- Book chapter
Broy, M.: Software engineering — from auxiliary to key technologies. In: Broy, M., Denert, E. (eds.) *Software Pioneers*, pp. 10–13. Springer, Heidelberg (2002)
- Online document
Cartwright, J.: Big stars have weather too. IOP Publishing PhysicsWeb.
<http://physicsweb.org/articles/news/11/6/16/1> (2007). Accessed 26 June 2007

Always use the standard abbreviation of a journal's name according to the ISSN List of Title Word Abbreviations, see

- [ISSN.org LTWA](http://www.issn.org/LTWA)

If you are unsure, please use the full journal title.

For authors using EndNote, Springer provides an output style that supports the formatting of in-text citations and reference list.

- [EndNote style \(zip, 2 kB\)](#)

TABLES

- All tables are to be numbered using Arabic numerals.
- Tables should always be cited in text in consecutive numerical order.
- For each table, please supply a table caption (title) explaining the components of the table.
- Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.
- Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.

ARTWORK AND ILLUSTRATIONS GUIDELINES

Electronic Figure Submission

- Supply all figures electronically.
- Indicate what graphics program was used to create the artwork.
- For vector graphics, the preferred format is EPS; for halftones, please use TIFF format. MSOffice files are also acceptable.
- Vector graphics containing fonts must have the fonts embedded in the files.
- Name your figure files with "Fig" and the figure number, e.g., Fig1.eps.

Line Art

- Definition: Black and white graphic with no shading.

- Do not use faint lines and/or lettering and check that all lines and lettering within the figures are legible at final size.
- All lines should be at least 0.1 mm (0.3 pt) wide.
- Scanned line drawings and line drawings in bitmap format should have a minimum resolution of 1200 dpi.
- Vector graphics containing fonts must have the fonts embedded in the files.

Halftone Art

- Definition: Photographs, drawings, or paintings with fine shading, etc.
- If any magnification is used in the photographs, indicate this by using scale bars within the figures themselves.
- Halftones should have a minimum resolution of 300 dpi.

Combination Art

- Definition: a combination of halftone and line art, e.g., halftones containing line drawing, extensive lettering, color diagrams, etc.
- Combination artwork should have a minimum resolution of 600 dpi.

Color Art

- Color art is free of charge for online publication.
- If black and white will be shown in the print version, make sure that the main information will still be visible. Many colors are not distinguishable from one another when converted to black and white. A simple way to check this is to make a xerographic copy to see if the necessary distinctions between the different colors are still apparent.
- If the figures will be printed in black and white, do not refer to color in the captions.
- Color illustrations should be submitted as RGB (8 bits per channel).

Figure Lettering

- To add lettering, it is best to use Helvetica or Arial (sans serif fonts).
- Keep lettering consistently sized throughout your final-sized artwork, usually about 2–3 mm (8–12 pt).
- Variance of type size within an illustration should be minimal, e.g., do not use 8-pt type on an axis and 20-pt type for the axis label.
- Avoid effects such as shading, outline letters, etc.
- Do not include titles or captions within your illustrations.

Figure Numbering

- All figures are to be numbered using Arabic numerals.
- Figures should always be cited in text in consecutive numerical order.
- Figure parts should be denoted by lowercase letters (a, b, c, etc.).
- If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures, "A1, A2, A3, etc." Figures in online appendices (Electronic Supplementary Material) should, however, be numbered separately.

Figure Captions

- Each figure should have a concise caption describing accurately what the figure depicts. Include the captions in the text file of the manuscript, not in the figure file.

- Figure captions begin with the term Fig. in bold type, followed by the figure number, also in bold type.
- No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.
- Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.
- Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.

Figure Placement and Size

- Figures should be submitted separately from the text, if possible.
- When preparing your figures, size figures to fit in the column width.
- For most journals the figures should be 39 mm, 84 mm, 129 mm, or 174 mm wide and not higher than 234 mm.
- For books and book-sized journals, the figures should be 80 mm or 122 mm wide and not higher than 198 mm.

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If you include figures that have already been published elsewhere, you must obtain permission from the copyright owner(s) for both the print and online format. Please be aware that some publishers do not grant electronic rights for free and that Springer will not be able to refund any costs that may have occurred to receive these permissions. In such cases, material from other sources should be used.

Accessibility

In order to give people of all abilities and disabilities access to the content of your figures, please make sure that

- All figures have descriptive captions (blind users could then use a text-to-speech software or a text-to-Braille hardware)
- Patterns are used instead of or in addition to colors for conveying information (colorblind users would then be able to distinguish the visual elements)
- Any figure lettering has a contrast ratio of at least 4.5:1

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Springer accepts electronic multimedia files (animations, movies, audio, etc.) and other supplementary files to be published online along with an article or a book chapter. This feature can add dimension to the author's article, as certain information cannot be printed or is more convenient in electronic form. Before submitting research datasets as electronic supplementary material, authors should read the journal's Research data policy. We encourage research data to be archived in data repositories wherever possible.

Submission

- Supply all supplementary material in standard file formats.
- Please include in each file the following information: article title, journal name, author names; affiliation and e-mail address of the corresponding author.
- To accommodate user downloads, please keep in mind that larger-sized files may require very long download times and that some users may experience other problems during downloading.

Audio, Video, and Animations

- Aspect ratio: 16:9 or 4:3
- Maximum file size: 25 GB
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Text and Presentations

- Submit your material in PDF format; .doc or .ppt files are not suitable for long-term viability.
- A collection of figures may also be combined in a PDF file.

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- Spreadsheets should be submitted as .csv or .xlsx files (MS Excel).

Specialized Formats

- Specialized format such as .pdb (chemical), .wrl (VRML), .nb (Mathematica notebook), and .tex can also be supplied.

Collecting Multiple Files

- It is possible to collect multiple files in a .zip or .gz file.

Numbering

- If supplying any supplementary material, the text must make specific mention of the material as a citation, similar to that of figures and tables.
- Refer to the supplementary files as “Online Resource”, e.g., “... as shown in the animation (Online Resource 3)”, “... additional data are given in Online Resource 4”.
- Name the files consecutively, e.g. “ESM_3.mpg”, “ESM_4.pdf”.

Captions

- For each supplementary material, please supply a concise caption describing the content of the file.

Processing of supplementary files

- Electronic supplementary material will be published as received from the author without any conversion, editing, or reformatting.

Accessibility

In order to give people of all abilities and disabilities access to the content of your supplementary files, please make sure that

- The manuscript contains a descriptive caption for each supplementary material
- Video files do not contain anything that flashes more than three times per second (so that users prone to seizures caused by such effects are not put at risk)

Appendix C1. Types of Sentences in a Social Story™

Sentence Types (Gray, 2004)		Updated Sentence Types (Gray, 2015)	
Descriptive	Factual, identify key aspects of a situation or topic, answer “wh” questions, bring “logic and accuracy” to a social story.	Descriptive	Accurately describe context and gives meaning to other sentences, including cultural beliefs/norms. Can describe observable external factors as well as internal factors such as another person’s thoughts/feelings.
Perspective	Refer to another person’s internal thoughts, feelings, beliefs and wellbeing.	Coaching:	Guide behaviour through describing possible responses by the child and caregivers.
Cooperative	Highlights what other people will do to help the child in a situation.	Audience	Describing possible responses/choices by the child in a situation that are likely to be effective or expected.
Directive	Identify possible responses that the child could make in order to guide them in the situation.	Team	Describe possible ways that the caregiver can respond to support the individual.
Affirmative	Used to emphasise an important point, reassure the child or refer to a rule.	Self-coaching	A coaching sentence written by the child themselves.
Control	Sentences to help a child with autism put the social story into practice and remember it. These are written by the child themselves.		

Appendix C2. Measures for the Social Stories Project

Autism Spectrum Disorder-Behaviour Problems for Children (ASD-BPC; Matson and Gonzalez, 2007)

Please read the list of behaviours below and rate how frequently your child shows this behaviour.

Frequency: 0=Never, 1=Almost Never, 2=Seldom, 3=Half the Time, 4=Usually, 5=Almost Always, 6=Always

Then for behaviours that do occur (with a rating of 1 or above), please rate how intense you think the behaviour is.

Intensity: 1= low, 2=mildly intense, 3=moderately intense, 4= really intense, 5=maximally intense.

Behaviour	How frequently does this behaviour occur (0-6)	How intense is this behaviour usually (1-5)
1. Poking him/her self in the eye		
2. Harming self by hitting, pinching, scratching, etc.		
3. Kicking objects		
4. Mouthing or swallowing objects causing bodily harm		
5. Removal of clothing at inappropriate times		
6. Unusual play with objects		
7. Inappropriate sexual behaviour		
8. Playing with own saliva		
9. Throwing objects at others		
10. Banging on objects with hand		
11. Smearing or playing with feces (poo)		
12. Leaving the supervision of caregiver without permission		
13. Aggression towards others		
14. Pulling others' hair		
15. Yelling or shouting at others		
16. Property destruction		
17. Repeated and unusual vocalizations.		
18. Repeated and unusual body movements		
19. If there is a behaviour not listed please write it here:		

School will develop a Social Story to address ONE of these behaviours. This will be called the target behaviour.

Motivation Assessment Scale (MAS; Durand, 1986)

Please describe the target challenging behaviour on the lines below in as much detail as possible. Describe each physical movement that makes up the challenging behaviour from start to finish in the order in which it happens. Describe exactly what we would expect to see during the challenging behaviour. Please include a description of how you would know when the behaviour has started and stopped. Make sure you specify which part of the body is involved and whether any contact with another object or person would be made.

Example of challenging behaviour: Hitting.

The behaviour begins when the child raises either arm into the air with their fist clenched and directs it towards an object or another person. The child moves their arm in a downward motion with force towards the object/person. The behaviour stops when the arm returns to a downward position with the fist down. The child is likely to make contact with the object/person, although it is possible that contact will not be made if the other person moves away. In this situation, the behaviour would still be classified as hitting if the other criteria were met.

Now please read each question below carefully and circle the one number that best describes your observations of this behaviour.

0=Never, 1=Almost Never, 2=Seldom, 3=Half the Time, 4=Usually, 5=Almost Always, 6=Always

	Rating (0-6)
1. Would the behaviour occur continuously, over and over, if this person were left alone for long periods of time? (For example, several hours)	
2. Does the behaviour occur following a request to perform a difficult task?	
3. Does the behaviour seem to occur in response to you talking to other persons in the room?	
4. Does the behaviour ever occur to get a toy, food, or activity that this person has been told that he or she can't have?	

5. Would the behaviour occur repeatedly, in the same way, for very long periods of time, if no one were around? (For example, rocking back and forth for over an hour.)	
6. Does the behaviour occur when any request is made of this person?	
7. Does the behaviour occur whenever you stop attending to this person?	
8. Does the behaviour occur when you take away a favourite toy, food, or activity?	
9. Does it appear to you that this person enjoys performing the behaviour? (It feels, tastes, looks, smells, and/or sounds pleasing.)	
10. Does this person seem to do the behaviour to upset or annoy you when you are trying to get him or her to do what you ask?	
11. Does this person seem to do the behaviour to upset or annoy you when you are not paying attention to him or her? (For example, if you are sitting in a separate room, interacting with another person.)	
12. Does the behaviour stop occurring shortly after you give this person the toy, food, or activity he or she has requested?	
13. When the behaviour is occurring, does this person seem calm and unaware of anything else going on around him or her?	
14. Does the behaviour stop occurring shortly after (one to five minutes) you stop working or making demands of this person?	
15. Does this person seem to do the behaviour to get you to spend some time with him or her?	
16. Does the behaviour seem to occur when this person has been told that he or she can't do something he or she had wanted to do?	

Social Skills Questionnaire Adapted – Teacher*

Date: _____ Pupil's name: _____
 His/Her sex: _____ School: _____
 His/Her age: _____ Teacher's name: _____

Please put a circle around the rating which best describes this pupil over the past week.

Circle the number 0 if the item is not true. Circle the number 1 if the item is sometimes true. Circle the number 2 if the item is mostly true.

Please answer all items.

No.	Question	Not true	Sometimes true	Mostly true
1	Listens to other people's points of view during arguments	0	1	2
2	Makes requests from teachers in a polite way	0	1	2
3	Controls his/her temper when he/she loses in a game or competition	0	1	2
4	Reacts appropriately if peers tease him/her or say unkind things	0	1	2
5	Asks to join in activities with peers in an appropriate manner	0	1	2
6	Expresses affection or positive feelings to others	0	1	2
7	Does kind things for others voluntarily	0	1	2
8	Gives compliments or says nice things to others when appropriate	0	1	2
9	Controls his/her temper when told off or criticized by teachers	0	1	2
10	Asks permission before borrowing or using other people's things	0	1	2
11	Shares things with peers	0	1	2
12	Controls his/her temper during disagreements with peers	0	1	2
13	Asks peers if he/she may join in their activities	0	1	2
14	Has an appropriate facial expression (e.g. not excessive grinning or aggressive)	0	1	2
15	Apologizes when he/she does something wrong	0	1	2
16	Spends free time in the company of peers	0	1	2
17	Invites others to join in games or activities	0	1	2
18	Tells a teacher if he/she has a problem or needs help	0	1	2
19	Expresses sympathy or concern to others who are hurt or upset	0	1	2
20	Follows the rules in games or activities	0	1	2
21	Takes part in games and activities with peers	0	1	2
22	Takes part in conversations with adults	0	1	2

Name of young person: _____

Teacher: _____

23	Makes eye contact appropriately with others during conversations	0	1	2
24	His/her tone of voice is appropriate (e.g. not aggressive or unusual)	0	1	2
25	Controls his/her temper when he/she does not get his/her own way with teachers	0	1	2
26	Laughs or smiles when appropriate	0	1	2
27	Takes part in conversations with peers	0	1	2
28	Shows that he/she is listening to others during conversations	0	1	2
29	Can express his/her feelings of anger but without losing his/her temper	0	1	2
30	Stands up for him/herself appropriately if peers act unreasonably	0	1	2

*Adapted from the Social Skills Questionnaire-Teacher (SSQ-T): Spence, S. H. (1995). Social skills training : Enhancing social competence with children and adolescents. Windsor, U.K.: NFER-Nelson

Behaviour Recording Before Social Story

Day 1

MORNING			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

AFTERNOON			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

Day 2

MORNING			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

AFTERNOON			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

Behaviour Recording After Social Story

Day 1

MORNING			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

AFTERNOON			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

Day 2

MORNING			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

AFTERNOON			
Time of day	Behaviour seen? Tick one box per episode	How long did it last? (minutes)	Severity (1-5)
Before break			
After break			

Young Person Feedback Form

1. How helpful did you find the social story that you were read?

Please put a mark on the line below to let us know how you feel.

Helpfulness scale



The social story was
not helpful for me.

Don't know



The social story was
helpful for me.

2. In what ways did the social story help?

3. What could have been better?

4. Would it be helpful for your teacher to keep reading you the social story?

Circle your answer below.

No

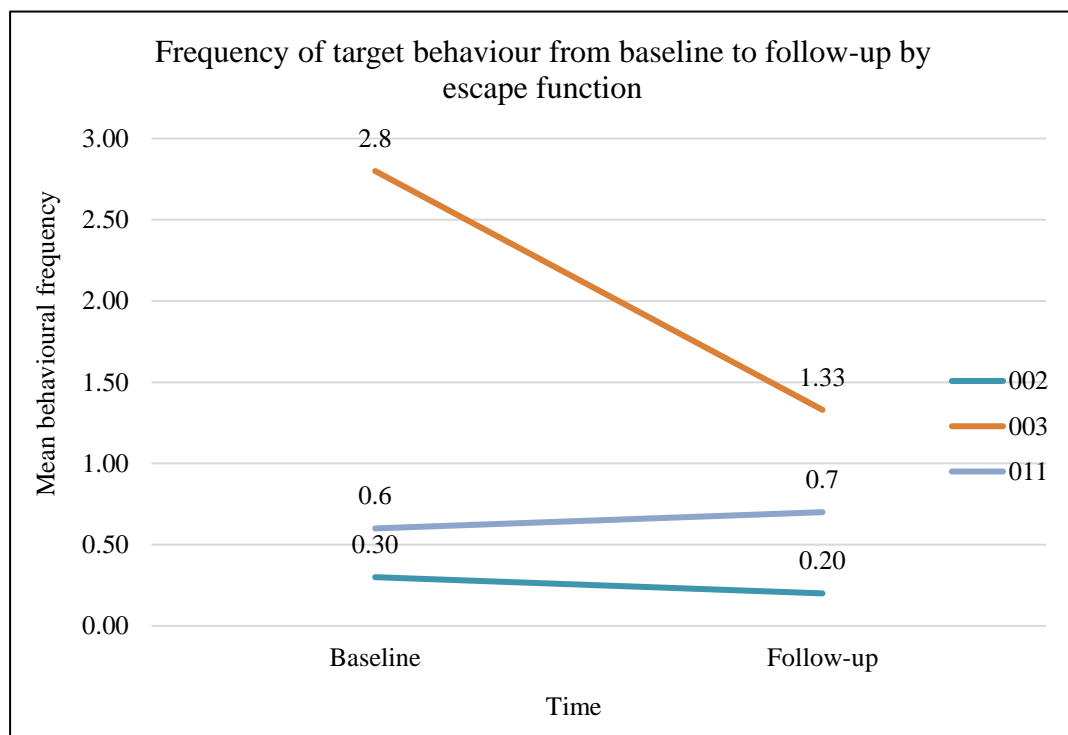
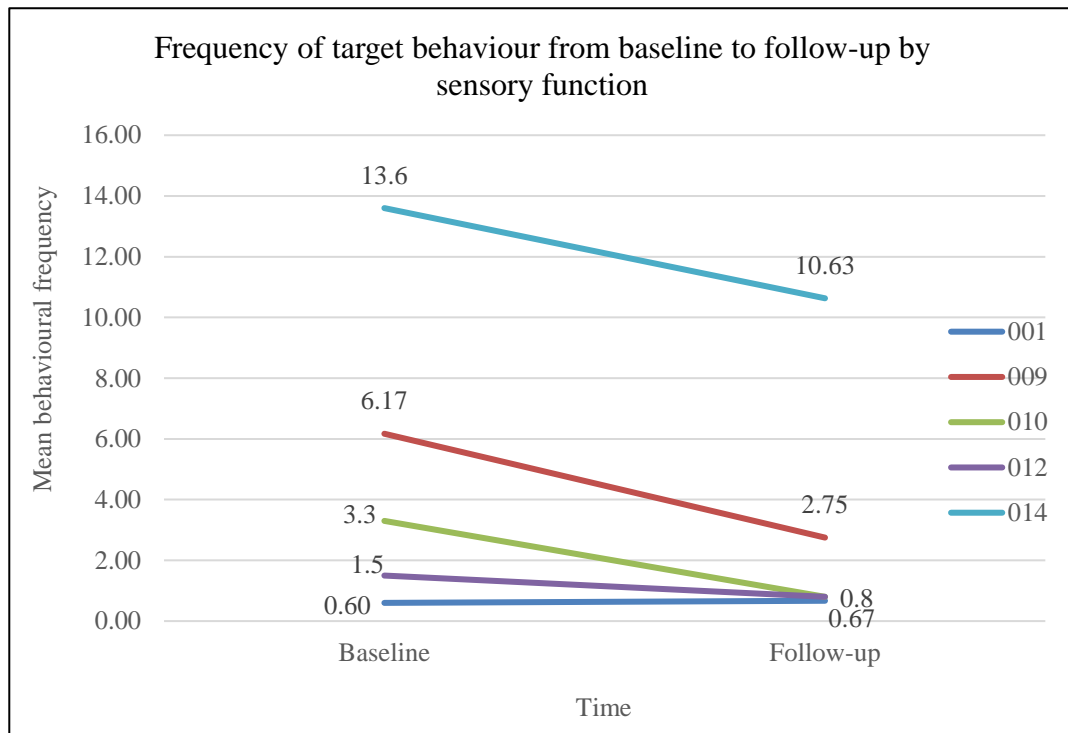
Don't
know

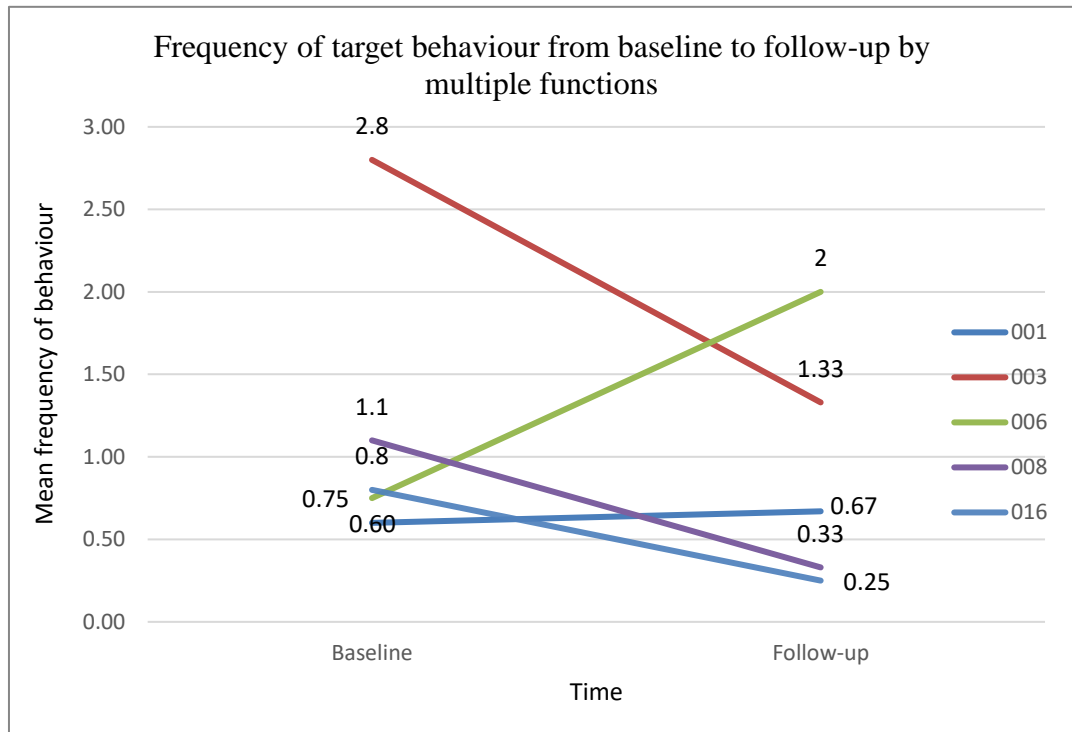
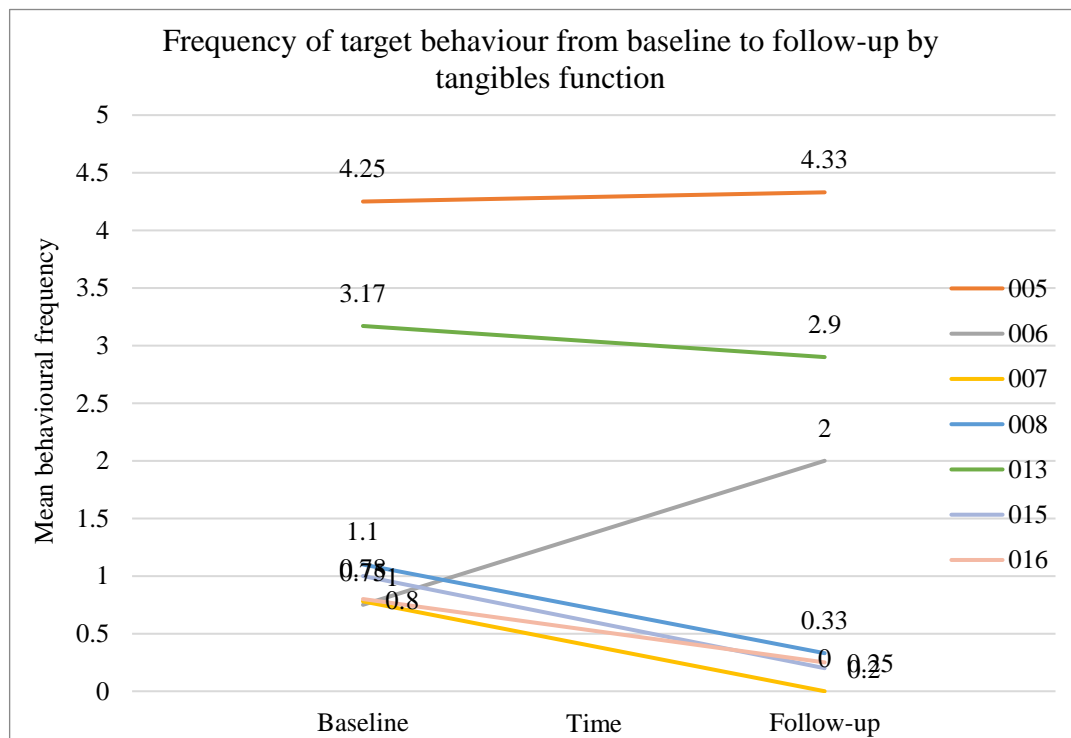
Yes

Appendix C3. Case Series Summary Table of Statistics by Participant

Participant code	Age	Gender	TR Frequency (0=TR-; 1=TR+)	Young Person's Feedback Form?	% change in frequency of behaviour	Rank change in frequency (1=biggest reduction)	Function 1	Function 2	FC? (0=FC-; 1=FC+)	SSQ TR	SSQ % change	Rank SSQ change	Any violation sentences (0=yes; 1=no)?	No. of violation sentences	Social story ratio	Comprehension level	Rank comprehension level
001	6	M	0	1	11.67%	13	1	4	1	0	-11.1%	11	0	6	1	P7	7.5
002	5	M	1	1	-33.33%	9	2		1	0	-16.0%	12	0	2	3	P6	10.5
003	7	M	1	1	-52.50%	7	2	3	1	nc	nc	nc	1	0	1	1c	5.5
004	6	M	nc	1	nc	nc	1	3	1	nc	nc	nc	1	0	1.5	1c	5.5
005	10	F	0	1 teacher	1.88%	12	4		0	1	125.0%	1	0	2	1	P6	10.5
006	10	M	0	1 teacher	166.67%	15	4	3	1	0	-44.0%	14	0	1	3	1A	2
007	9	M	1	0	-100.00%	1	4		0	1	82.4%	3	0	2	2	P5.6	13.5
008	9	F	1	0	-70.00%	4	4	1,2,3	nc	1	111.8%	2	0	4	0	P6	10.5
009	9	M	1	0	-55.43%	6	1		1	0	9.1%	6	1	0	1.2	1B	3.5
010	13	M	1	1	-75.76%	3	1		0	0	0.0%	9.5	0	2	1.5	P4	15
011	12	M	0	0	16.67%	14	2		nc	1	28.6%	4	0	1	1	P5	13.5
012	6	M	1	0	-46.67%	8	1		1	0	18.2%	5	0	1	2	P3i	16
013	7	F	0	0	-8.52%	11	4		1	0	6.7%	7	0	3	1	P6	10.5
014	11	M	0	0	-21.84%	10	1		0	0	0.0%	9.5	0	2	0	2B	1
015	9	M	1	0	-80.00%	2	4		0	0	-23.3%	13	0	2	0.5	P7	7.5
016	9	M	1	1	-68.75%	5	4	1	1	0	4.5%	8	0	2	0.5	1B	3.5

Appendix C4. Graphs of Treatment Response by Function






Appendix C5. SPSS Table of Correlations between Proportion of Sentence Types and Magnitude (%) of Change in Frequency of Behaviour

Correlations		Prop descript	Prop coach	Prop viol	Freq change
Prop_descript	Pearson Correlation	1	.243	-.801**	.224
	Sig. (2-tailed)		.382	.000	.422
	N	15	15	15	15
Prop_coach	Pearson Correlation	.243	1	-.759**	-.252
	Sig. (2-tailed)	.382		.001	.365
	N	15	15	15	15
Prop_viol	Pearson Correlation	-.801**	-.759**	1	-.030
	Sig. (2-tailed)	.000	.001		.916
	N	15	15	15	15
Freq_change	Pearson Correlation	.224	-.252	-.030	1
	Sig. (2-tailed)	.422	.365	.916	
	N	15	15	15	15


** . Correlation is significant at the 0.01 level (2-tailed).

Appendix C6. Ethical Approval from the University of Bath Psychology Ethics Committee



psychology-ethics | Emily Norris

Re: Amendment for chair's approval Ref: 16-071

 You replied to this message on 15/07/2016 15:38.

15/07/2016

MessageHeaderAnalyzer

+ Get more add-ins

Dear Emily,

Thank you for notifying us of this potential amendment to your study. This seems reasonable and does not raise new ethical concerns, and thus is approved by Chair's Action.

Best wishes,
Dr Michael Proulx

From: Emily Norris
Sent: Monday, July 11, 2016 5:54 PM
To: psychology-ethics
Subject: Amendment for chair's approval Ref: 16-071

Dear Dr Proulx,

Reference Number 16-071: The relationship between challenging behaviour and Social Story Interventions: an observational study in a naturalistic setting

In my original ethics application, I stated that the exclusion criteria for my study would be a specified cut-off level of comprehension for young people with autism. Having spoken with the schools involved in the study, I have discovered that many of them use social stories with young people with a wide range of comprehension, therefore the proposed cut-off would lose a large proportion of the sample.

I am therefore requesting chair's approval for an amendment to my ethics application that comprehension will no longer be an exclusion criteria; all children with a diagnosis of autism will be included regardless of comprehension, with the only proviso being that the children fall within the remit that the school would usually write a social story for. However, comprehension will be included as a variable during analysis to determine if this has any effect on outcome.

Best wishes,

Emily Norris
Clinical Psychologist in Training
University of Bath



Dear Emily,

Yes, that's fine. I am happy to approve that amendment via Chair's Action. Please could you email your ethics application with that amendment highlighted for our records?

Please could you also confirm that you have an up-to-date DBS certificate to work with children.

Best of luck with your data collection,

Dr. Nathalia Gjersoe
Ethics Committee Chair

From: Emily Norris

Sent: 23 September 2016 09:24

To: psychology-ethics <psychology-ethics@bath.ac.uk>

Subject: Re: Amendment Reference Number 16-071 Emily Norris

Dear Nathalia,

Reference Number 16-071: The relationship between challenging behaviour and Social Story Interventions: an observational study in a naturalistic setting

Thank you for your response. The rationale for the original proposal was that an adult over the age of 16 who was able to demonstrate capacity to make a decision and consent would not require parental permission in other decision-making processes, such as healthcare. As such, the protocol was designed to follow the same process in order to respect the views of the individual with autism as an adult with capacity.

However, I appreciate that in this situation it may not be comfortable to override the wishes of the parents when the individual is still in full-time education. I will amend my testing procedure to state that both the parent and individual need to consent in order for them to take part. Would it be possible for you to confirm that I have approval to proceed with 16-19 year olds based on this change please?

Best wishes,

Emily

Emily Norris
Clinical Psychologist in Training
University of Bath

From: psychology-ethics
Sent: 19 September 2016 15:40
To: Emily Norris
Subject: RE: Amendment Reference Number 16-071 Emily Norris
Dear Emily,

I have considered this amendment with another member of the Committee. The move to recruit participants from 16-19 is not a problem, participants from 16 years up are able to give consent for participation in studies. However we do not think it is appropriate to over-ride a parent's stated preference that their child not take part in the study. Please could you amend so that testing proceeds only if consent has been received from BOTH parent and the individual. Let me know if you feel this will overly constrain your recruitment and we can discuss.

All the best,

Dr. Nathalia Gjersoe
Ethics Committee Chair

From: Emily Norris
Sent: 26 August 2016 13:04
To: psychology-ethics <psychology-ethics@bath.ac.uk>
Cc: Ailsa Russell <A.J.Russell@bath.ac.uk>
Subject: Amendment Reference Number 16-071 Emily Norris
Dear Dr Proulx,
Reference Number 16-071: The relationship between challenging behaviour and Social Story Interventions: an observational study in a naturalistic setting
Ethical amendment
I am requesting an amendment please by chair's action to extend the age range for recruitment to the above study. Currently I have approval to recruit up to age 16. However, in some secondary special school settings, young people remain on in full-time education up to the age of 19, rather than moving on to college settings. I am therefore requesting to extend the recruitment age to include young people age 16-19 years who remain in full-time secondary education.

For young people aged 16-19 years, consent would still be sought from parents as they remain in full time education. However, the assent form has been modified and would now become a consent form for this age bracket (see attached). As such, if a parent has consented but a young person in this age bracket does not give consent, the young person will be assumed to have capacity to consent and will **not** be included in the study. If both parents and the young adult give consent they **will** be included. If the young adult aged 16-19yrs gives consent, but their parent/guardian does not give consent, a capacity assessment will take place to answer the question: does this young adult have capacity to **make a decision** to take part in the specified research study without needing parental permission?

According to the Mental Capacity Act (MCA, 2005), there are two stages to consider when assessing capacity:
1. "Does the person have an impairment, or a disturbance in the functioning, of their mind or brain? This can include, for example, conditions associated with mental illness, concussion, or symptoms of drug or alcohol abuse.
2. Does the impairment or disturbance mean that the person is unable to make a specific decision when they need to? You should offer all appropriate and practical support to achieve this before applying this stage of the test."
(Mental Capacity Act, 2005).

Participants with a learning disability taking part in this study would fulfil the first stage as having "impairment, or a disturbance in the functioning, or their mind or brain".

The second stage would require an assessment covering the following points as outlined in the MCA:

- Is the young adult able to understand the decision to be made (whether to take part in the specified research study or not) using the information provided on the information sheet?
- Is the person able to retain the information given about the study?
- Is the young adult able to use the information provided to weigh up the pros and cons of taking part?
- Is the young person able to communicate the decision that they have made

If the outcome of the assessment indicates that the young person has capacity to make the decision to take part, they **will** be included in the study. If they do not have capacity, they will **not** be included in the study as their parent has not consented.

I look forward to hearing back. I have attached the amended forms for young adults age 16-19yrs to this e-mail.

Please let me know if any further information is required to consider this amendment.

Best wishes,

Emily Norris



Dear Emily,

Thank you for alerting us to this amendment. I am happy to confirm that you have full ethical approval.

Best of luck with your data collection,

Dr. Nathalia Gjersoe
Chair, Psychology Ethics Committee

From: Emily Norris
Sent: 03 April 2017 14:24
To: psychology-ethics <psychology-ethics@bath.ac.uk>
Subject: Reference Number 16-071: Request for Amendment via Chair's Action

Dear Nathalia,

Reference Number 16-071: The relationship between challenging behaviour and Social Story Interventions: an observational study in a naturalistic setting

I am requesting an amendment via chair's action please for a minor addition to a debrief form following a revision in methodology. The study originally planned to send the same two questionnaires to teachers and parents to gather additional information about behaviour, and this was therefore stated on the information sheet. However, due to practicalities of sending questionnaires to parents which would have introduced additional difficulties to recruitment including delays, it was decided that it was not necessary for parents to complete the questionnaire as the research was interested in behaviour occurring within the school setting, not at home, therefore the questionnaires would not provide any new information. As such, questionnaires were not sent home apart from one school.

I have therefore added a brief paragraph to the debrief form to explain this, and was planning to send the original debrief form to the one school where parental questionnaires were obtained. I have highlighted the new paragraph on the attached document and would appreciate if this might be considered for approval via chair's action please.

Thank you,

Emily Norris
Clinical Psychologist in Training
University of Bath

Appendix C7. Ethics Documents for Educational Staff

The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Participant Information Sheet – Educational Staff VI.

Your school has agreed to take part in the following piece of research. This will involve various members of educational staff in the ways outlined below. We would like to invite you to be part of this study. Before you decide, it is important to understand why this is being done and what it will involve. Please read this information carefully. You will need to sign to say that you have understood this information before you can continue. By signing this form, you are consenting to take part in the study.

Why is this study being done?

We are hoping to find out more about the way in which challenging behaviour can impact on the usefulness of social stories.

A social story is a story developed by a parent/teacher/support worker with a young person. The story aims to describe a social situation or skill and how a child might respond best in that situation in order to develop social skills. Your school already uses social stories regularly to help children, and you may be familiar with them or have had experience with using them in school.

Challenging behaviour is a term often used to describe behaviour which has an impact on a young person's ability to access everyday activities, or behaviour which causes harm to themselves or others.

Social Stories have been found to be helpful in reducing challenging behaviour in a proportion of young people. However it is not yet fully understood which features of a social story might be particularly helpful. This study aims to find out if there are certain aspects of social stories that help with certain types of challenging behaviour.

Why have we been chosen and do we have to take part?

Your school has agreed to take part in the study, and we are therefore inviting members of educational staff at the school to take part, including yourself. However, you do not have to take part in the study. If you decide to take part and then later change your mind, you can stop taking part at any point without giving your reasons, and, if you wish, your data from the questionnaires will be destroyed.

What will we be asked to do if we take part?

Different members of staff will be asked to do different tasks within the research procedure. Not all of the tasks below may apply to you. This will be decided with the school once the research begins. The researchers will not be collecting any information about you personally; you will only be asked to complete questionnaires and collect data relating to children at the school.

If you agree to take part, you may be asked to complete several questionnaires about behaviours shown by several children at your school which you would describe as 'challenging'. Parents will also be given these questionnaires. Although multiple behaviours shown by the children might be described as challenging, the questionnaires will be used to identify and prioritise one challenging behaviour per child that occurs in the educational setting. This behaviour will be the focus for the remainder of the study.

We may ask you to keep a record of the children's challenging behaviour for up to 1 week. If you are asked to do this, you will be given a behavioural record form to complete which involves

marking how long the behaviour lasts and rating how intense the behaviour was during that time. We will ask you to make a note of this each time you see the behaviour during the school day. The researcher will show staff involved in this part of the study how to use the behaviour record. The researcher may also carry out some additional observations of the behaviour in the classroom setting for the purposes of reliability ratings.

If you are familiar with writing social stories, you may be asked to write a social story to help with the identified behaviour. This should be written in exactly the same way as you would normally write it. If applicable, you would then be asked to read the social story to the relevant child twice a day over a one to two-week period at school. The social story will be made available to the researchers so they can examine the features that might particularly relate to the challenging behaviour.

At the end of this period, if you completed the questionnaires at the beginning of the study, you will be asked to complete the same questionnaires again. We may ask you to keep a record of the challenging behaviour once again for up to a week using the same record as before. Researchers will use this information to find out if there have been any changes in the challenging behaviour.

Will my experiences and reports be kept confidential?

Yes, all information which is collected from you during the course of the research will be kept confidential within the research team. It will conform to the Data Protection Act of 1998 with respect to data collection, storage and destruction. This means that all paper-based and electronic information will be locked and password protected with access restricted to the research team. Any information that you complete about challenging behaviour will have your name removed so that you cannot be identified from it.

We hope to report our findings in academic/health related journals and present them to relevant health professionals at meetings and conferences. The findings will also contribute to Emily Norris' Doctorate in Clinical Psychology. You will not be identified in any reports or publications arising from the study.

Are there any advantages/benefits from taking part?

It is hoped that the social story will help children and staff at the school to better understand and manage social situations which trigger their challenging behaviour. However, as every child is different, social stories may not be effective for every young person. We hope that the information we collect from you about the children in your school may help to improve our understanding of how social stories can help when children and young people show behaviour that challenges. The school will also be offered compensation for taking part in the study which will go towards an identified need in the school.

Are there any disadvantages/risks from taking part?

We don't consider there to be any disadvantages to taking part. It is possible that you may not see any improvement in the children's behaviour during this brief study, and in some cases the behaviour may worsen whilst the new intervention is tried. You are free to stop taking part in the study at any time, as are the children and parents at the school. If you notice any signs that a child does not wish to continue in the study, please inform the researchers. It is important for you to understand that you are not required to disclose anything that you do not want to and you should disclose only the things which you feel are relevant.

What if there is a problem?

If you have any concerns or wish to complain about any aspect of the way you have been approached or treated as part of this study, you should first contact the researchers who will do their best to answer your questions. Their contact details are provided at the end of this information form and again on the debriefing form at the end of the survey. If you remain unhappy and wish to complain

formally, you can do this by contacting the University of Bath Psychology Ethics committee at psychology-ethics@bath.ac.uk.

What to do next if I'm interested?

If you would like to take part, please read the consent form and respond to the statements, then sign at the bottom to show that you have read it and agree to consent. If you have any further questions or would like to speak to a researcher before deciding whether to take part, you can contact the research team using the details below.

Emily Norris
Clinical Psychologist in
Training
E-mail: en325@bath.ac.uk

Dr Ailsa Russell
Clinical Supervisor
E-mail: a.j.russell@bath.ac.uk

Dr Mark Brosnan
Field Supervisor
E-mail: m.j.brosnan@bath.ac.uk

Consent Form – Educational Staff

CONFIDENTIAL

Title of project: The relationship between challenging behaviour and Social Story interventions:
an observational study in a naturalistic setting

Name of Primary Researcher: Emily Norris

Please circle:

1. I have read the information sheet about this study Yes / No
2. I have had the opportunity to ask questions and discuss this study Yes / No
3. I have received satisfactory information about this study Yes / No
4. I understand I am free to withdraw from this study: Yes / No
 - At any time
 - Without giving a reason for withdrawing
 - And that I can omit questions on the questionnaire that I do not wish to answer
5. I agree to take part in this research Yes / No

Signed

Date

Name in Block Letters:

Signature of researcher

Date

The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Debriefing Form Educational Staff V.1

Thank you for taking part in this study with your school.

The primary aim of this research was to learn more about factors that might influence how effective a social story is for a young person with autism. In particular, we wanted to find out how important it is to know the reason behind behaviour before writing a social story in order for it to be effective and whether social stories are more helpful for certain types of behaviour than others. To answer this question, we compared the function of challenging behaviour to the function targeted in the social story to see how closely they matched, and how successful the outcome was of that social story. We were also interested in whether social stories have an effect on a young person's social skills, and whether the types of sentences used in social stories affects how helpful they are. Your data will help us to learn more about ways to manage challenging behaviour, and how to improve social stories in the future so that they can be helpful for more young people with autism.

Once again results of this study will not include your name or any other identifying characteristics. The research did not use deception. You may have a printed copy of this summary if you wish, and you may also request a summary of the research findings once the project is completed if you wish.

If you have any questions or concerns following this research, please contact the primary researcher Emily Norris on the details below, or another member of the research team (Dr Ailsa Russell or Dr Mark Brosnan). If you wish to complain formally about any aspect of the way you have been approached or treated as part of this study, you can do this by contacting the University of Bath Psychology Ethics committee at psychology-ethics@bath.ac.uk.

For further information about the topics in the research, the National Autistic Society offers some helpful information on challenging behaviour in ASD: <http://www.autism.org.uk/about/behaviour/challenging-behaviour.aspx>. Further information about social stories can be found at the following website created by the founder of social stories: <http://carolgraysocialstories.com/>

Thank you for your participation in this research.

Contact details for the research team:

Emily Norris
Clinical Psychologist in
Training
E-mail: en325@bath.ac.uk

Dr Ailsa Russell
Clinical Supervisor
E-mail:
a.j.russell@bath.ac.uk

Dr Mark Brosnan
Field Supervisor
E-mail:
m.j.brosnan@bath.ac.uk

Key references:

For a review of evidence into social stories and future research, see: Kokina, A. & Kaczmarek, L.A., (2013). *Social Story interventions for children with Autism Spectrum Disorders: Current evidence and unanswered questions*. In V.B. Patel, V. Preedy, & C. Martin (Eds.), *The Comprehensive Guide to Autism*. Dordrecht, Heidelberg, London, New York: Springer.

Appendix C8. Ethics Documents for Parents

The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Participant Information Sheet – Parents V1.

You are invited to take part in the following piece of research with your child. Before you decide, it is important to understand why this is being done and what it will involve. Please read this information carefully. You will need to sign to say that you have understood this information before you can continue. By signing this form, you are consenting to yourself and your child participating in this research.

Why is this study being done?

We are hoping to find out more about the way in which challenging behaviour can impact on the usefulness of social stories.

A social story is a story developed by a parent/teacher/support worker with a young person. The story aims to describe a social situation or skill and how a child might respond best in that situation in order to develop social skills. Your child's school uses social stories regularly to help children.

Challenging behaviour is a term often used to describe behaviour which has an impact on a young person's ability to access everyday activities, or behaviour which causes harm to themselves or others.

Social Stories have been found to be helpful in reducing challenging behaviour in a proportion of young people. However it is not yet fully understood which features of a social story might be particularly helpful. This study aims to find out if there are certain aspects of social stories that help with certain types of challenging behaviour.

Do I have to take part?

No, you do not have to take part in the study. If you and your child decide to take part and then later change your minds, you can stop taking part at any point without giving your reasons, and, if you wish, your data will be destroyed.

What will we be asked to do if we take part?

If you agree to take part, you will be asked to complete several questionnaires about a behaviour shown by your child which you would describe as 'challenging'. Members of staff at the school will also be given these questionnaires. Although several behaviours shown by your child might be described as challenging, the questionnaires will be used to identify and prioritise one challenging behaviour that occurs in the educational setting. This behaviour will be the focus for the remainder of the study.

The behaviour will be 'measured' in terms of its frequency and intensity for approximately 1 week. The researcher may also carry out some additional observations of the behaviour in the classroom setting for the purposes of reliability ratings.

Your child's teacher will then write a social story to help with the identified behaviour. This will be read to your child every day over a one to two-week period at school. The

social story will be made available to the researchers so they can examine the features that might particularly relate to the challenging behaviour.

At the end of this period, you and the teaching staff will be asked to complete the same questionnaires. Your child will be asked some questions about how helpful they found the social story. The challenging behaviour in the classroom will be measured again for 1 week. Researchers will use this information to find out if there have been any changes in the challenging behaviour.

Will my experiences and reports be kept confidential?

Yes, all information which is collected about you and your child during the course of the research will be kept confidential within the research team. It will conform to the Data Protection Act of 1998 with respect to data collection, storage and destruction. This means that all paper-based and electronic information will be locked and password protected with access restricted to the research team. Any information about you will have yours and your child's name removed so that you cannot be identified from it.

We hope to report our findings in academic/health related journals and present them to relevant health professionals at meetings and conferences. The findings will also contribute to Emily Norris' Doctorate in Clinical Psychology. You will not be identified in any reports or publications arising from the study.

Are there any advantages/benefits from taking part?

It is hoped that the social story will help your child to better understand and manage social situations which trigger their challenging behaviour. However, as every child is different, social stories may not be effective for every young person. We hope that the information we collect from you and other participants may help to improve our understanding of how social stories can help when children and young people show behaviour that challenges. The school will also be offered compensation for taking part in the study which will go towards an identified need in the school.

Are there any disadvantages/risks from taking part?

We don't consider there to be any disadvantages to taking part. It is possible that you may not see any improvement in your child's behaviour during this brief study, and in some cases the behaviour may worsen whilst the new intervention is tried. You are free to stop taking part in the study at any time, for example if you become worried about changes in your child's behaviour. It is important for you to understand that you are not required to disclose anything that you do not want to and you should disclose only the things which you feel are relevant.

What if there is a problem?

If you have any concerns or wish to complain about any aspect of the way you have been approached or treated as part of this study, you should first contact the researchers who will do their best to answer your questions. Their contact details are provided at the end of this information form and again on the debriefing form at the end of the survey. If you remain unhappy and wish to complain formally, you can do this by contacting the University of Bath Psychology Ethics committee at psychology-ethics@bath.ac.uk.

What to do next if I'm interested?

If you would like to take part, please read the consent forms and respond to the statements, then sign at the bottom to show that you have read it and agree to consent. If you have any further questions or would like to speak to a researcher before deciding whether to take part, you can contact the research team using the details below.

Emily Norris
Clinical Psychologist in
Training
E-mail: en325@bath.ac.uk

Dr Ailsa Russell
Clinical Supervisor
E-mail: a.j.russell@bath.ac.uk

Dr Mark Brosnan
Field Supervisor
E-mail: m.j.brosnan@bath.ac.uk

Consent Form - Parents

CONFIDENTIAL

Title of project: The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Name of Primary Researcher: Emily Norris

Please circle:

1. I have read the information sheet about this study Yes / No
2. I have had the opportunity to ask questions and discuss this study Yes / No
3. I have received satisfactory information about this study Yes / No
4. I understand I am free to withdraw from this study: Yes / No
 - At any time
 - Without giving a reason for withdrawing
 - And that I can omit questions on the questionnaire that I do not wish to answer
5. Do you agree to take part by:

Completing the questionnaires? Yes / No
 Your child participating in the social story project? Yes / No

Signed

Date

Name in Block Letters:

Signature of researcher

Date

The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Debriefing Form Parents V.1 (questionnaires)

Thank you for taking part in this study with your son/daughter.

The primary aim of this research was to learn more about factors that might influence how effective a social story is for a young person with autism. In particular, we wanted to find out how important it is to know the reason behind behaviour before writing a social story in order for it to be effective and whether social stories are more helpful for certain types of behaviour than others. To answer this question, we compared the function of challenging behaviour to the function targeted in the social story to see how closely they matched, and how successful the outcome was of that social story. We were also interested in whether social stories have an effect on a young person's social skills, and whether the types of sentences used in social stories affects how helpful they are. Your data will help us to learn more about ways to manage challenging behaviour, and how to improve social stories in the future so that they can be helpful for more young people with autism.

Once again results of this study will not include your name or any other identifying characteristics. The research did not use deception. You may have a printed copy of this summary if you wish, and you may also request a summary of the research findings once the project is completed if you wish.

If you have any questions or concerns following this research, please contact the primary researcher Emily Norris on the details below, or another member of the research team (Dr Ailsa Russell or Dr Mark Brosnan). If you wish to complain formally about any aspect of the way you have been approached or treated as part of this study, you can do this by contacting the University of Bath Psychology Ethics committee at psychology-ethics@bath.ac.uk.

For further information about the topics in the research, the National Autistic Society offers some helpful information on challenging behaviour in ASD: <http://www.autism.org.uk/about/behaviour/challenging-behaviour.aspx>. Further information about social stories can be found at the following website created by the founder of social stories: <http://carolgraysocialstories.com/>

Thank you for your participation in this research.

Contact Details for the research team:

Emily Norris
Clinical Psychologist in
Training
E-mail: en325@bath.ac.uk

Dr Ailsa Russell
Clinical Supervisor
E-mail: a.j.russell@bath.ac.uk

Dr Mark Brosnan
Field Supervisor
E-mail: m.j.brosnan@bath.ac.uk

Key references:

For a review of evidence into social stories and future research, see: Kokina, A. & Kaczmarek, L.A., (2013). *Social Story interventions for children with Autism Spectrum Disorders: Current evidence and unanswered questions*. In V.B. Patel, V. Preedy, & C. Martin (Eds.), *The Comprehensive Guide to Autism*. Dordrecht, Heidelberg, London, New York: Springer.

The relationship between challenging behaviour and Social Story interventions: an observational study in a naturalistic setting

Debriefing Form Parents V.2 (no questionnaires)

Thank you for taking part in this study with your son/daughter.

The primary aim of this research was to learn more about factors that might influence how effective a social story is for a young person with autism. In particular, we wanted to find out how important it is to know the reason behind behaviour before writing a social story in order for it to be effective and whether social stories are more helpful for certain types of behaviour than others. To answer this question, we compared the function of challenging behaviour to the function targeted in the social story to see how closely they matched, and how successful the outcome was of that social story. We were also interested in whether social stories have an effect on a young person's social skills, and whether the types of sentences used in social stories affects how helpful they are. Your data will help us to learn more about ways to manage challenging behaviour, and how to improve social stories in the future so that they can be helpful for more young people with autism.

Whilst we had originally planned to collect questionnaire data from parents as well as teachers, this was not included as part of the final procedure for your school. All observations and social story interventions were targeted towards school, therefore we only required questionnaire data specific to this setting.

Once again results of this study will not include your name or any other identifying characteristics. The research did not use deception. You may have a printed copy of this summary if you wish, and you may also request a summary of the research findings once the project is completed if you wish.

If you have any questions or concerns following this research, please contact the primary researcher Emily Norris on the details below, or another member of the research team (Dr Ailsa Russell or Dr Mark Brosnan). If you wish to complain formally about any aspect of the way you have been approached or treated as part of this study, you can do this by contacting the University of Bath Psychology Ethics committee at psychology-ethics@bath.ac.uk.

For further information about the topics in the research, the National Autistic Society offers some helpful information on challenging behaviour in ASD: <http://www.autism.org.uk/about/behaviour/challenging-behaviour.aspx>. Further information about social stories can be found at the following website created by the founder of social stories: <http://carolgraysocialstories.com/>

Thank you for your participation in this research.

Contact Details for the research team:

Emily Norris
Clinical Psychologist in
Training
E-mail: en325@bath.ac.uk

Dr Ailsa Russell
Clinical Supervisor
E-mail:
a.j.russell@bath.ac.uk

Dr Mark Brosnan
Field Supervisor
E-mail:
m.j.brosnan@bath.ac.uk

Key references:

For a review of evidence into social stories and future research, see: Kokina, A. & Kaczmarek, L.A., (2013). *Social Story interventions for children with Autism Spectrum Disorders: Current evidence and unanswered questions*. In V.B. Patel, V. Preedy, & C. Martin (Eds.), *The Comprehensive Guide to Autism*. Dordrecht, Heidelberg, London, New York: Springer.

Reading age:
11-13 years

Participant Information Sheet Young Person:
Challenging Behaviour and Social Stories

We are hoping to do some research with young people with autism and would like to invite you to take part.

What is research?

Research is a way of trying to learn more about a topic and find the answers to some questions.

Why are we doing this research?

Sometimes, children and young people with autism can behave in ways that harm themselves or others, or that gets in the way of everyday activities.



This is often called 'challenging behaviour'.

Why do young people behave in ways we find challenging?

There are lots of reasons why young people might show this behaviour. For example, it might be a way of telling people how they are feeling or what they need.



How can we help?



One way that we can help young people with their difficult behaviour is through using social stories.

We want to find out if social stories are more helpful in certain situations and for certain types of challenging behaviour.



What is a social story?

Social stories are stories that describe a social situation or skill.



The story tells a young person what might happen in a situation, how it might make them feel and helpful ways that they could respond.

Do I have to take part?

No. You can choose whether you want to take part or not. We will also ask your parents if they are happy for you to take part.



I've changed my mind...

You can stop taking part in the research at **any time** by telling the researcher, the teacher or your parent/guardian.

It is okay to do this and you will not get in trouble if you choose to stop taking part.

What will I be asked to do?



Your parents and teachers will be asked some questions about a behaviour that is challenging.



Your teachers will keep a record of the behaviour.



They will then write you a social story about the behaviour and read this to you each day for about two weeks.



After this, your teachers will keep a record again of the behaviour. They may still read you the social story.



Your parents and teachers will be asked the same questions as at the start of the study and you will be asked some questions too.



What will happen to the information?

Researchers will use information they collect to learn more about challenging behaviour and social stories.

The information that you give us will be kept safe and your name will be taken off it. Only the researchers will know whose information is whose.



Once we have done this, the information collected in the research may be written up into a report and shared with others involved or interested in our findings.

Assent Form Young Person

What should I do if I want to take part?

Your parents have already given permission for you to take part in the study however we want to know that you are happy with this too.

If you agree to take part, please put a tick next to each sentence below and write your name on the line.

- I have read the information about the research/
An adult has explained it to me ☐
- I have had the chance to ask questions ☐
- I know that I can stop taking part at any time ☐
- I agree to take part in the study and am happy to be asked
some questions, for my behaviour to be observed and for
my teachers to read me a social story. ☐

.....
Name of young person

.....
Signature of supporting adult

.....
Name of supporting adult

Reading age:
11-13 years

Debriefing Form Young Person:
Challenging Behaviour and Social Stories



Thank you for taking part!

The main question in our research was:

How can we make social stories more helpful for young people with autism?



We tried to answer this by measuring your behaviour before and after a social story.

We also looked at the reasons behind behaviour that can be seen as challenging.

By taking part in our study, you have helped us to learn more about how to help young people with their challenging behaviour through social stories.



Your information will be kept safe and your name will be taken off it.



If you have any questions or worries about the research, please talk to your parents or teachers who may be able to help, or to the research team.

Thank you!



Peer review

This journal operates a single blind review process. All contributions will be initially assessed by the editor for suitability for the journal. Papers deemed suitable are then typically sent to a minimum of two independent expert reviewers to assess the scientific quality of the paper. The Editor is responsible for the final decision regarding acceptance or rejection of articles. The Editor's decision is final. [More information on types of peer review](#).

Use of word processing software

It is important that the file be saved in the native format of the word processor used. The text should be in single-column format. Keep the layout of the text as simple as possible. Most formatting codes will be removed and replaced on processing the article. In particular, do not use the word processor's options to justify text or to hyphenate words. However, do use bold face, italics, subscripts, superscripts etc. When preparing tables, if you are using a table grid, use only one grid for each individual table and not a grid for each row. If no grid is used, use tabs, not spaces, to align columns. The electronic text should be prepared in a way very similar to that of conventional manuscripts (see also the [Guide to Publishing with Elsevier](#)). Note that source files of figures, tables and text graphics will be required whether or not you embed your figures in the text. See also the section on Electronic artwork.

To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Article structure

Article formatting and style

Formatting and style in the text should follow the style used by the American Psychological Association. You are referred to the Publication Manual of the American Psychological Association, Sixth Edition, ISBN 978-1-4338-0561-5, copies of which may be ordered

from <http://books.apa.org/books.cfm?id=4200067> or APA Order Dept., P.O.B. 2710, Hyattsville, MD 20784, USA or APA, 3 Henrietta Street, London, WC3E 8LU, UK.

Font, headings, and other requirements stipulated should be adhered to.

Word Count

The maximum word count for articles submitted to the journal is 8,000 words, including references and tables.

Subdivision - numbered sections

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